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Editorial

Migration and Healthcare services

Sri Lanka has a long and proud history of providing high-quality healthcare services to its citizens. However, the recent migration of healthcare workers, including doctors, nurses, and trained paramedical staff, to other countries poses a significant threat to the future of our nation's healthcare.

One of the immediate consequences of healthcare worker migration is the shortage of skilled professionals in the country's healthcare sector. As doctors, nurses, and paramedical staff leave for better opportunities abroad, the local healthcare system is struggling to maintain an adequate workforce. This shortage places immense pressure on the remaining healthcare workers, leading to increased workloads and burnout.
The shortage of healthcare workers directly affects the quality and accessibility of healthcare services for the Sri Lankan population. Longer waiting times, reduced access to specialized care, and overburdened healthcare facilities are becoming commonplace. This will lead to compromised health outcomes, increased morbidity, and mortality rates. This shortage also places a strain on the country's healthcare infrastructure. Overcrowded hospitals and clinics, coupled with limited resources, make it challenging to provide adequate care to patients.

A strong healthcare system relies on skilled professionals, research, and development. When healthcare workers leave the country, Sri Lanka loses valuable human resources and expertise that are essential for the advancement of its healthcare system. The lack of skilled professionals hinders the implementation of innovative medical technologies and practices, preventing the healthcare system from evolving and meeting the changing needs of the population.

Another worrying effect of the current exodus of healthcare workers is its impact on the training and development of the next generation of healthcare professionals. With fewer teachers, mentors and role models, aspiring doctors, nurses, and paramedical staff will face difficulties in acquiring the necessary skills and knowledge to excel in their careers. This lack of training and deterioration in standards will have severe long-term consequences for the quality of healthcare in Sri Lanka.

To address these challenges, the Sri Lankan government and healthcare officials must first recognize the importance of retaining its healthcare workforce and investing in healthcare education and infrastructure.

The age old saying "Health is wealth" is most applicable to our country today than ever before. While the government and policy makers pay attention to increasing the monetary wealth of our country, health appears to be of secondary concern. A far-sighted vision is required to understand the consequences of poor healthcare on the development of a nation. One of the basic necessities of a prosperous nation is a healthy population, without which all hopes for a developed future will remain a dream.

Chief editor
Dr G .R. Francis

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Original research Papers

An audit on the prevalence of malignancy from reported cases of TH Batticaloa; A hospital- based retrospective analysis.

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1. Teaching Hospital, Batticaloa.

Background:

A retrospective analysis of reported malignant cases from Teaching Hospital Batticaloa from January 2017 to December 2022 was conducted to know the sociodemographic profile, cancer site, and histological type among the patients.

Introduction and Objectives:

Cancers are one of the major health problems among people worldwide. Their incidence among the general population in Sri Lanka is also on a continuous rise. So that knowledge about sociodemographic and histopathological profiles of malignant cases is important in identifying target groups for screening and implementing preventive measures, we have conducted a retrospective analysis on reported histopathological cases from our hospital.

Methods:

Histopathological reports of 2428 patients diagnosed with various types of cancers from the year 2017 to 2022 were analyzed for a sociodemographic profile, cancer site, and histological type. SPSS 21 software was used for statistical analysis.

Results:

According to our analysis, more women (50.1%) were diagnosed with cancers than men. Most malignant cases reported belong to the age group of 60-64 years (n=403, 16.6%). The most common cancer site was lip, tongue, and mouth (20.7%) followed by breast (10.3%). Squamous cell carcinoma was the most common histopathological type (41.1%) in both genders. The major cancer site in females was the breast (n= 244) whereas lip tongue and mouth were the most common in males (n=301). Cancers which are lip tongue & mouth, oesophagus, and lung are more prevalent in 65-69 years, colorectal and cervix are more prevalent in 60-64 years, bladder and prostate are more prevalent in 70-74 years, breast and ovary are more prevalent in 50-54 years and thyroid is more prevalent in 35-39 years.

Though cancer cases were almost constant from 2017 to 2019, there was a drop in 2020 & 2021 in the total number of reported cases probably due to COVID-19 lockdown restrictions followed by a significant rise in 2022 (n=515, 21.2%).

Conclusion:

This study highlights that breast and oral cancer burdens are still higher than other types and the need to reevaluate the adequacy of screening measures for these cancer types to have a good outcome.

References

1. 22nd Publication – National Cancer Incidence and Mortality Data Sri Lanka: 2020. Published in March 2023
2. Moujhuri Nandi, Abhijit Mandal, Anupam Kumar Asthana (2013). Audit of Cancer Patients from Eastern Uttar Pradesh (UP), India: A University Hospital Based Two Year Retrospective Analysis. Asian Pac J Cancer Prev, 14 (9), 4993-4998
3. Das BP (2005). Cancer pattern in haryana: twenty-one years' experience (a retrospective analysis of 15968 patients). Health Administrator, 1052, 29-49.
4. Jemal A, Bray F, Center MM, et al (2011). Global cancer statistics. CA Cancer J Clin, 61, 69-90.
5. Siegel R, Naishadham D, Jemal A (2013). Cancer Statistics, 2013. Ca Cancer J Clin, 63, 11-3.

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Original research Papers

Assessment of quality of life among end stage renal disease patients undergoing hemodialysis at Teaching Hospital batticaloa, Sri Lanka

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Abstract:

Introduction:

End Stage Renal Disease (ESRD) patients confront several challenges in their life with the introduction of routine hemodialysis treatment. However, there is limited data on the Quality of Life (QOL) of ESRD patients undergoing hemodialysis in Sri Lanka.

Objective:

To assess the QOL among ESRD patients undergoing Hemodialysis in Teaching Hospital Batticaloa (THB), Sri Lanka.

Methods:

A descriptive cross-sectional study was conducted on ESRD patients undergoing hemodialysis at THB. Data was collected through an interviewer-administered questionnaire which gathered sociodemographic and clinical information, along with the WHOQOL-BREF questionnaire measuring QOL under the following four domains: physical, psychological, social relationships and, environmental domains. Descriptive statistics were employed for data analysis. Ethical approval was obtained prior to conducting the study.

Results:

More than half of the participants experienced poor QOL under each of the four domains. The highest mean QOL score was observed in the social relationships domain (57.36±3.895), while the physical domain (41.78±3.187) had the lowest mean QOL score.

Discussion:

The findings indicate that the majority of ESRD patients (75.56%) undergoing hemodialysis at THB, have poor overall QOL. The obtained results highlight the need to address the QOL issues faced by these patients through targeted support and interventions.

Introduction

Renal Replacement Therapy, encompassing renal transplantation and dialysis, has significantly enhanced and prolonged the lives of individuals with End Stage Kidney Disease (ESKD), who were on the verge of death(1). Renal Transplantation is considered the best management option for ESKD due to its irreversible nature but is often constrained by the high cost and scarcity of kidneys(2). In instances where Renal Transplantation is awaited, or not possible, dialysis, which includes hemodialysis and peritoneal dialysis, acts as a life-sustaining therapy for ESKD patients by mechanically filtering waste products and excess fluid from the body (3). The global prevalence of dialysis patients surpassed 2 million in 2010, and projections suggest that this number will more than double by 2030(4).

However, with the introduction of routine hemodialysis treatments, patients with ESKD confront several changes in their life(5). They become dependent on the dialysis procedure, requiring regular visits to dialysis centers, and adhering to strict treatment schedules. Additionally, patients face numerous physiological stressors such

as fluid and dietary restrictions, reduced mobility, lethargy, and potential complications associated with therapy and vascular access surgery(5)(6)(7). These alterations can have a profound impact on their relationships and overall independence(1).

Given these challenges, assessing the Quality of Life (QOL) of ESKD patients undergoing hemodialysis is of utmost importance. It is crucial to consider patients' priorities to optimize their well-being alongside clinical outcomes(8). However, there is a dearth of data regarding QOL among ESKD patients undergoing hemodialysis within the Sri Lankan context. As the global demand for hemodialysis continues to rise, gaining insights into the QOL of these patients is essential for healthcare practitioners, policymakers, and families to better understand and address their needs.

The objective of this study is to assess the QOL among ESRD patients undergoing Hemodialysis in THB, the only Teaching hospital in the Eastern province. By evaluating multiple QOL domains, this research will provide insights into the challenges faced by ESKD patients and help formulate targeted interventions.

Methods

A Cross-Sectional Descriptive study was conducted at the Hemodialysis Unit THB among the patients with ESKD undergoing hemodialysis at THB. The complete enumeration method was used as the sampling method, including all eligible patients during the period of January to March 2022. Data collection was performed using an interviewer-administered questionnaire consisting of three sections.

Section A and Section B of the questionnaire focused on gathering socio-demographic information and clinical details, respectively. Socio-demographic information included factors such as age, gender, ethnicity, marital status, employment status, educational background, and monthly household income. Clinical details encompassed the frequency and duration of hemodialysis treatment and the comorbidities present.

Section C of the questionnaire utilized the World Health Organization Quality of Life-BREF (WHOQOL-BREF) questionnaire, which assessed the quality of life across four domains: Physical domain, psychological domain, social relationships domain, and environmental domain. Each domain was assessed using a Likert scale. The collected data was analyzed using the Statistical Package for the Social Sciences (SPSS) version 23. The mean scores of the items within each domain were calculated to obtain the raw scores, which were subsequently transformed into a 0-100 scale score using a transformation formula provided in the WHOQOL-BREF user manual. The overall quality of life was determined based on the participants' scores across all four domains.

To categorize the quality of life, a cutoff value was determined. Participants with a score below 60 were categorized as having a "poor QOL," while those with a score of 60 or higher were categorized as having a "good QOL." This cutoff value was chosen based on previous research (9), which demonstrated a sensitivity of 95% and excellent negative predictive value in identifying individuals with probable worse quality of life.

Ethical approval was obtained from the Ethics Review Committee, Faculty of Health Care Sciences, Eastern University, Sri Lanka. Informed consent was obtained from each participant prior to their inclusion in the study.

Results

Out of the 56 patients registered in the Hemodialysis unit for maintenance Hemodialysis, a total of 45 participants who met the inclusion criteria were recruited for the study.

Sociodemographic details of Participants

Table 1 Sociodemographic details of the participants

Objective	Categories	Frequency	Percentage (%)
Age	21-40	15	33.33
	41-60	28	62.2
	>60	2	4.4
Gender	Male	27	60.0
	Female	18	40.0
Marital status	Single	4	8.9
	Married	35	77.8
	Widowed	4	8.9

Ethnicity	Tamil	30	66.7
	Muslim	8	17.8
Highest education received	Grade 5 or below	6	13.3
	Grade 6-10	18	40.0
	O/L	9	20.0
	A/L	6	13.3
	Tertiary education	6	13.3
Employment status	Full time	9	20.0
	Unemployed	30	66.7
Monthly household income	<20.000 Rs	27	60.0
	20,001 Rs to 40,000 Rs	9	20.0
	>40,000 Rs	9	20.0

The study population included 27 males and 18 females, with a male-to-female ratio of 3:2. The majority of male participants were in the age group of 41-50 years, while most female participants were in the age group of 51-60 years.

Regarding ethnicity, 66.7% of the participants were of Tamil ethnicity. The majority of the participants were married (77.8%), and 62.2% were between 41-60 years of age. Approximately 60% of the participants had a monthly income of less than 20,000 Sri Lankan Rupees. Of the participants, 20% had full-time employment, while 66.67% were unemployed and 13.33% worked part-time.

Clinical details of Participants

In terms of comorbidities, 86.7% of the participants had hypertension, making it the most prevalent comorbidity among ESKD patients undergoing hemodialysis at THB. Diabetes Mellitus was the second most common comorbidity, affecting 37.8% of the participants. Regarding the duration of hemodialysis, 62.2% of the participants had been undergoing treatment for less than 18 months, while 11% had been on hemodialysis for more than 3 years. The majority of participants (97%) underwent hemodialysis twice per week.

Quality of Life (QOL) of Participants

Among the domains, the social relationships domain had the highest mean value of 57.36±3.895, indicating relatively better QOL in this aspect. In contrast, the physical domain had the lowest mean value of 41.78±3.187, indicating comparatively poorer QOL in terms of physical well-being.

	Mean		Std. Deviation
	Statistic	Std. Error	Statistic
Physical Domain	41.78	3.187	21.382
Psychological Domain	47.29	2.639	17.700
Social Relationships Domain	57.36	3.895	26.127
Environmental Domain	55.62	2.299	15.425

In the Physical Domain, a majority of participants (75.55%) relied on medical treatment for daily functioning, while 60% reported limited energy for day-to-day activities. 68.89% of participants were dissatisfied/very dissatisfied with their sleep.

Within the Psychological Domain, approximately half of the participants (23 participants-51.11%) felt that they were not able to concentrate at all or concentrate only in a little amount. 5 (11.11%) participants were almost always having negative feelings such as blue mood, despair, anxiety, and depression while majority of the participants had those feelings very often (19 participants-42.22%).

In the Social Relationships domain, a majority of participants (71.11%) expressed satisfaction with their personal relationships, while 48.89% felt satisfied with the support they received from their friends. However, 33.34% of participants reported dissatisfaction or significant dissatisfaction with their sexual life.

Within the Environmental Domain, 24.45% of participants expressed dissatisfaction or significant

dissatisfaction with their home environment. Additionally, financial constraints were evident, as 22.22% of participants reported not having enough financial resources to meet their needs, and 35.56% had only limited financial support. Furthermore, a significant portion (66.67%) of participants had no opportunities for leisure activities. On a positive note, the accessibility for healthcare was satisfactory for 73.33% of participants, and the transport facility to and from the hospital was highly satisfactory for 53.33% of participants. However, 11.11% expressed significant dissatisfaction with transportation.

According to the results of the study, only 24.44% had a QOL equal to or above 60 while 75.56% had a QOL score below 60 (Poor QOL)

Comparison of overall QOL with demographic data

Non-parametric tests were used in the bivariate when determining the association between demographic factors and clinical factors with overall QOL. A p value of less than 0.05 was considered statistically significant. Marital Status and Monthly household income were found to be statistically significant with the overall QOL of the participants.

Overall QOL of Participants

Table 3 Comparison of Overall QOL with demographic data

Compare group	Significance level
Overall QOL vs Age	0.694
Overall QOL vs Gender	0.675
Overall QOL vs Marital Status	0.027
Overall QOL vs Ethnicity	0.975
Overall QOL vs Highest Education received	0.107
Overall QOL vs Employment status	0.484
Overall QOL vs Monthly household income	0.004

Discussion

Our study examined the sociodemographic and clinical characteristics and quality of life (QOL) outcomes of patients undergoing hemodialysis at the Hemodialysis Unit of THB.

In terms of sociodemographic characteristics, our study showed that the majority of participants were males (60%), of Tamil ethnicity (66.7%), married (77.8%), aged between 41-60 years (62.2%), had completed education up to grade 6-10 (40%), were unemployed (66.7%), had a monthly income less than 20,000 Sri Lankan Rupees (60%), and were undergoing hemodialysis for less than 6 months (31.1%). These sociodemographic factors are in line with another study conducted in Sri Lanka, involving National Hospital Sri Lanka (NHSL) and Teaching Hospital Anuradhapura (THA) (10), which reported similar characteristics, except for the ethnicity factor where most participants were from Sinhala ethnicity (79.6%). Additionally, a study conducted in Indonesia in 2022 also showed comparable results, with a majority of male participants (67.2%), aged 35-50 years (40.2%), and having middle-level education (73.8%) (11).

Regarding QOL outcomes, our study found that a significant proportion of participants experienced poor QOL in the physical (86.67%), psychological (80%), social relationships (55.56%), and environmental (64.44%) domains. The highest QOL score was observed in the social relationships domain (44.44%), while the lowest score was found in the physical domain (13.33%), indicating poor QOL across all four domains. These findings are consistent with a similar study conducted in Sri Lanka, where poor QOL was also reported in all four domains with the least QOL score in the physical domain (18%) (10). However, this study found the highest QOL score (47.2%) in the environmental domain, followed by the psychological domain (35.2%). A study conducted in Indonesia (11) also demonstrated poor QOL in all four domains, with the highest QOL observed in the environmental domain.

Comparing individual components within the physical and psychological domains, our study's results were largely similar to those of the study conducted in NHSL and THA in 2019 (10). However, our study showed significantly better results in the social relationships domain, with a higher proportion of participants reporting satisfactory personal relationships (71.11%) compared to the previous study (38%). Similarly, a higher percentage

of participants in our study (42.22%) expressed satisfaction with their sexual activity, while the previous study reported a much lower satisfaction rate (4.4%).

In the environmental domain, our study revealed that only 15.55% of participants had adequate financial support, while a significant number (35.56%) had only a little financial support and 22.22% had no financial support at all to meet their needs. This is a noteworthy finding, contrasting with the study conducted by(10), which reported that only a total of 36.8% of participants had inadequate financial resources, which is comparatively better.

When considering the associations with QOL, our study identified statistical significance between QOL and marital status (p=0.027) and monthly household income (p=0.004). In the study conducted in NHSL and THA(10), statistical significance was found with education level, monthly income, and frequency of dialysis. Moreover, a study conducted in Athens, Greece, revealed that marital status and educational level significantly influenced the QOL of hemodialysis patients(12).

Despite some limitations, such as the relatively small sample size and potential confounding factors, the findings of our study have important implications for clinical practice and future research. The high prevalence of poor QOL highlights the need for interventions and strategies to improve the various domains of QOL in hemodialysis patients. Addressing financial support, enhancing social relationships, and providing appropriate psychological support may be key areas to focus on.

References

1. Saad MM, El Douaihy Y, Boumitri C, et al. Predictors of quality of life in patients with end-stage renal disease on hemodialysis. Int J Nephrol Renovasc Dis. 2015;8:119–23.
2. Abecassis M, Bartlett ST, Collins AJ, et al. Kidney transplantation as primary therapy for end-stage renal disease: a National Kidney Foundation/ Kidney Disease Outcomes Quality Initiative (NKF/ KDOQITM) conference. Clin J Am Soc Nephrol [Internet]. 2008/02/06. 2008 Mar;3(2):471–80. Available from: https://pubmed.ncbi.nlm.nih.gov/18256371
3. Ian W, Raine T, Wiles K, Anna Goodhart, et al. Oxford Handbook of Clinical Medicine. Tenth. Oxford University Press; 2017. 894 p.
4. Chan CT, Blankestijn PJ, Dember LM et al. Dialysis initiation, modality choice, access, and prescription: conclusions from a Kidney Disease: Improving Global Outcomes (KDIGO) Controversies Conference. Kidney Int. 2019;96(1):37–47.
5. Bapat U, Kedlaya P. Perceived illness intrusion among patients on hemodialysis. Saudi Journal of Kidney Diseases and Transplantation [Internet]. 2009 May 1;20(3):386–91. Available from: https://www.sjkdt.org/article.asp?issn=1319-2442
6. Gutiérrez-Peredo GB, Martins MTS, da Silva FA, et al. Functional dependence and the mental dimension of quality of life in Hemodialysis patients: the PROHEMO study. Health Qual Life Outcomes [Internet]. 2020;18(1):234. Available from: https://doi.org/10.1186/s12955-020-01464-3
7. Paulo UDS, Viviani K, Santos F, et al. How to cite Complete issue More information about this article DAILY LIFE OF PATIENTS WITH CHRONIC RENAL FAILURE RECEIVING. 2008;
8. Evangelidis N, Tong A, Manns B, et al. Developing a Set of Core Outcomes for Trials in Hemodialysis: An International Delphi Survey. Am J Kidney Dis. 2017 Oct;70(4):464–75.
9. Silva PAB, Soares SM, Santos JFG, Silva LB. Cut-off point for WHOQOL-bref as a measure of quality of life of older adults. Rev Saude Publica. 2014;48(3):390–7.
10. Premadasa M, Hulangamuwa H, Wijesooriya W, Amarasekara T. Quality of Life Among Patients With Chronic Kidney Disease Who are Undergoing Haemodialysis at Two Selected Teaching Hospitals in Sri Lanka. OUSL Journal. 2019 Dec 30;14:31.
11. Darsini D, Notobroto HB, Afiyah RK, et al. Quality of Life of Hemodialysis Patients during COVID-19 Pandemic in Gatoel Hospital, Mojokerto City. Open Access Maced J Med Sci. 2022;10(E):293–302.
12. Vasilopoulou C, Bourtsi E, Giaple S, et al. The Impact of Anxiety and Depression on the Quality of Life of Hemodialysis Patients. Glob J Health Sci. 2016;8(1):45–55.

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Original research Papers

Experience in establishing Community Paediatric services at Teaching Hospital Batticaloa in Batticaloa District.

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Introduction and Background

Community Paediatrics is still a new subspeciality in Sri Lanka, encompassing neurodevelopmental disorders(NDDS) neuro disabilities and addressing vulnerable children. Around 43% of Children under five years in low-income countries and low middle income countries (LMICs) have a risk of not reaching their potential for development. In comparison to many other nations in the region, Sri Lanka, a low-middle income country, has awesome neonatal and infant mortality statistics. Improved healthcare and technology have increased the survival rates of at-risk infants, resulting in a higher risk for neuro-developmental disorders such as cerebral palsy, autism spectrum disorder, learning disorders, and behaviour abnormalities. Delays in early detection of at-risk children in LMICs may lead to NDDs, negatively impacting health, nutrition, learning and employment opportunities. Community pediatric teams provide services to high-risk infants, children with neurodevelopmental delays, and those with safeguarding issues to maximise developmental capacity and improve the quality of life for children and their families.

Community paediatric services were established in December 2022 at Teaching Hospital Batticaloa. The services are provided by an Acting Consultant Community Paediatrician, Medical Officer, Nursing Officers, Speech and Language Therapist, Occupational Therapist, Physiotherapist, and Counselling Assistant officer from the Divisional Secretariat Office. This clinic is held one day in the child guidance clinic within the paediatric clinic complex and one day in the paediatric professorial unit clinic. Patients are seen in the paediatric wards on other days.

Early intervention clinic

Team members

CVI intervention

There are no dedicated buildings, permanent staff, or therapists for community pediatric services in Teaching Hospital Batticaloa. The clinic is staffed by medical officer, nursing officers, and therapists who are not designated to community paediatric services.

Objectives

To describe the service delivery and community needs for the community paediatric clinic in the Teaching Hospital Batticaloa at Batticaloa District.

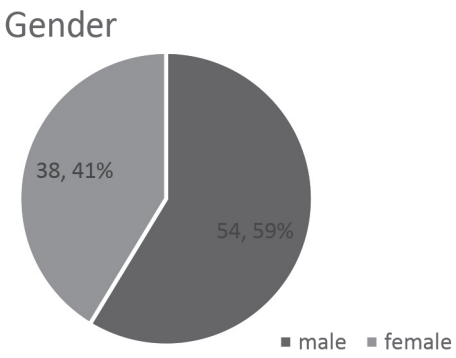
Method

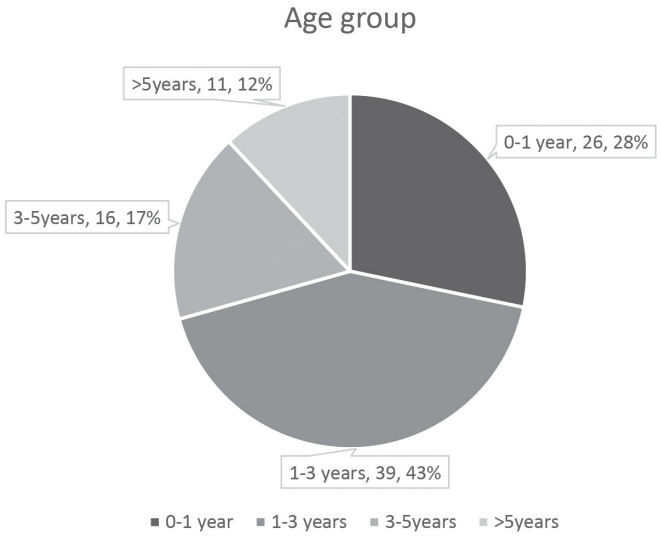
This is a retrospective clinical Audit. Data were collected from the clinic register of the Community Paediatric clinic in the Teaching Hospital Batticaloa in Batticaloa District from February 2023 to May 2023.

Results

The tertiary centre has received 92 referrals within four months. Of them, 54(59%) were males; 40(43%) were included 1-3 years of age. The majority were from the hospital paediatric unit (63/92, 68%) and neonatology unit (17/92, 18%). The clientele consisted mostly of Cerebral palsy (25/92, 27%), global developmental delay(24/92, 26%), high-risk neonates for Early intervention (15/92, 16%), Down syndrome (10/92, 11%), Autism Spectrum Disorder (9/92, 10%) learning difficulties (4/92, 4%) and other disorders (5/92, 5%). Among them, six children had cortical visual impairment (CVI) (6/92, 7%).

Demographic details





Referrals from	Total
Hospital paediatric units	61
Neonatal unit (THB)	17
Others	14

Discussion

Community Paediatrics is still a new subspeciality in paediatrics that involves understanding the complex interplay between physical, social, and environmental factors and human biology affecting the growth and development of all children and young people. Around 10% of Sri Lanka's child population requires specialised care due to various disabilities, developmental impairments, behavioural issues, and chronic conditions.

This audit's first aim is to identify the patients' categories, caseloads and where the referral comes from. Overall, the highest referrals came from hospital paediatric units; cerebral palsy accounted for the highest.

A similar Audit was done in Western Province in a secondary base hospital, and there were 33 referrals within two months duration. The majority were diagnosed with autism spectrum disorder.

Establishing a community paediatric centre in Low-middle-income countries is quite challenging. An ideal community pediatric clinic should be operated in a dedicated space with all facilities, including a sensory room. Children with neuro disabilities require lifelong support for their education, healthcare, social participation, and future employment. It should be provided holistically, acknowledging patients' and family members' unique challenges and needs. To offer patient-friendly services, it is essential to have a team of dedicated therapists and staff. Currently, Sri Lanka has Ayati, the National Centre for Children with Disabilities, which is situated in Ragama. It is an ideal model for community paediatrics services.

Conclusion

At a tertiary hospital level, there is still a high proportion of children presenting from the community with chronic neuro disabilities needing lifelong services.

Recommendations

Established community paediatric clinic centers with a multidisciplinary team are much needed in the Batticaloa district.

As there are no referrals from the community (by the Medical Officer of Health), awareness is needed regarding community paediatric clinic services in Teaching Hospital Batticaloa.

References

1. Walker SP, Wachs TD, Meeks Gardner J, Lozoff B, Wasserman GA, Pollitt E, et al. Child development: risk factors for adverse outcomes in developing countries. Lancet [Internet]. 2007 [cited 2023 Sep 17];369(9556):145–57. Available from: https://pubmed.ncbi.nlm.nih.gov/17223478/

2. Woythaler M. Neurodevelopmental outcomes of the late preterm infant. Semin Fetal Neonatal Med [Internet]. 2019 [cited 2023 Sep 17];24(1):54–9. Available from: https://pubmed.ncbi.nlm.nih.gov/30322826/

3. Vipulaguna D, Madhushani H a. H, Bandara RP. 1031 Establishing community paediatric services in Sri Lanka – translation of knowledge and skills from united kingdom to a lower- and middle-income country. Archives of Disease in Childhood [Internet]. 2022 Aug 1 [cited 2023 Sep 16];107(Suppl 2):A83–3. Available from: https://adc.bmj.com/content/107/Suppl_2/A83.1

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Original research Papers

Power and hierarchical positions & feedback practices at a medical school- An observational study

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Abstract:

Learner-centered feedback is one of the most powerful influences in learning and achievement. However specific literature on feedback for culturally diverse environments, such as Sri Lanka is lacking. By addressing this problem, we aimed to enhance learning and teaching in medical schools by identify and explore a culturally sensitive feedback guidance for Medical Education.

This qualitative, ethnographic, case study of the ethnically diverse Eastern University, Sri Lanka (EUSL) was emergent in design. Staff and students of EUSL represent predominantly three different ethnicities, Sinhala, Tamil and Muslim.

Data gathering utilized observation of learning conversations with three types of stakeholders, students, faculty and policy makers. Planned thematic analysis was usefully supplemented by discourse analysis in order to reduce uncritical imposition of western concepts.

Findings identified under-problematization of feedback practices, unaddressed cultural and historical complexities and willingness to develop post-colonial challenges to overly simplistic hegemonic "west is best" assumptions. Non-WEIRD (Westernised, Educated, Industrialised, Rich and Democratic) countries e.g. Sri Lanka can be benefitted from WEIRD evidence-based theories such as dialogic feedback and learner transitions best when informed by and adapted to local cultural practices and knowledge.

Key words:

Formative assessment, Learner-centered feedback, WEIRD, Cultural, Power, Hierarchy

Introduction

Formative assessment and feedback are characterized by differing conceptualisation such as dialogic, multi-sourced, incremental, self-evaluation and complicated by tensions between competing purposes, priorities and educational dominance by WEIRD countries.

Feedback may become more student-centred when students are empowered to self-regulate their learning. Sadler (1998) comments that it cannot be assumed, students will know what to do with "given feedback". However, Nicol & Macfarlane-Dick (2006) define self-regulated learning as an active constructive process whereby learners set goals, regulate and control their learning. Besides, teachers still tend to transmit feedback to learners by telling them what is right and what is wrong (Scager, Akkerman, Pilot, & Wubbels, 2017). So, the teacher needs to be sensitized as to how their feedback can be acted on and to ensure this is meaningful for the students (van Der Leeuw, Teunissen, & van Der Vleuten, 2018). For instance, teachers should understand or be aware that students can have different or changing ways of thinking such as fixed or growth mind-sets which influence their learning (Dweck, 2012). Again though it is noted this work was done in a western context.

"Correctives", where the teacher identifies errors in a student's performance and immediately delivers clarification (Guskey, 2010), and feedback are integral parts of normal ways of talking about

assessment (William, 2011). The seminal work on learning by Bloom (1964) in Guskey (2005) highlights individual differences between students and their use of assessment as a learning tool (Guskey, 2005; Guskey, 2007). However, William (2011) argues in a very important sense, that Bloom's distinction between "feedback" and "correctives" has been counter-productive and served to distort the original meaning of the term "feedback". Similarly, Ajjawi and Regehr (2019) suggest feedback gives information to students to understand where they are in their learning and what needs to be done to move their learning on. They encapsulate this with their recent definition of feedback, "a dynamic and co-constructive interaction in the context of a safe and mutually respectful relationship to challenge a learner's (and educators') ways of thinking, acting or being to support growth" (Ajjawi & Regehr, 2019, p. 653)

At the same time, several researchers have carried out cross-cultural, qualitative studies, albeit through a western lens, to better understand the study habits and conceptions of learning among students from different cultural backgrounds (e.g.Dahlin & Watkins, 2000; Marambe, Vermunt, & Boshuizen, 2012; Trompenaars, 2004). Assuming learner-centered feedback as one of the most powerful influences in learning and achievement (Bhattarai, 2007) there is a need to address the gap to help students learning in culturally diverse medical schools. Therefore, our aim for this study was to enhance learning and teaching in medical schools via identify and explore a culturally sensitive feedback guidance for Medical Education.

Methodology

Data collection was done using observation method as unobtrusively as possible to avoid any disturbance in the teaching-learning activities and to reduce the Hawthorne effect which is a type of reactivity in which participants in observations might modify their behaviour in response to their awareness of being observed (McCambridge, 2014). Observational data were analyzed using NVivo12 software to manage and categorize findings following multiple readings and manual initial coding. Though I used a predominantly AI method to collect and analyze data I was acknowledging the shadow with the lens of AI too as it would be wrong to deny some evidence of poor practices or negative inferences from the data (Patton,1999).

The observational findings are presented as data extracts from our field notes of nine observed learning conversations at FHCS, EUSL. The details of the observed learning conversation are given below in table 1

Table 1: Details of Observations

Obs	Place	Teaching Activity	Teacher	Also present	Students
1	Ward No 05, Teaching Hospital Batticaloa (THB)	Bedside teaching	Male- Tamil	4 doctors, 2 nurses,	20, Phase 2 students 9 female and 11 male
2	Tutorial room, THB	Discussion	Male- Tamil		20, Phase 2 students 9 female and 11 male
3	Clinical sciences lecture hall, THB	Lecture	Male- Tamil		19, Phase 2 students 8 male and 11 female
4	Surgery clinic THB	Group discussion	Male – Sinhala		23, Phase 3 students 11 male and 12 female
5	Surgical ward No 34 THB	Bedside teaching	Male- Tamil	3doctors, 1nurses, 18 patients	19, Phase 3 students 11 male 8 female
6	Lecture theatre @ FHCS	group discussion	Female- Tamil		16, Phase 1 students 3male 13 female
7	Pathophysiology Lecture hall@ FHCS	PBL review session	Four Teachers Tamil M2 F2		54, Phase 2 students 20 female and 34 male

Obs	Place	Teaching Activity	Teacher	Also present	Students
8	Auditorium FHCS	Students' Research presentation session	Twelve Teachers, Tamil M4F3 Sinhala M1F2 MuslimM1F1		8, Phase 2 students 100 students as audience
9	Ward No 04, THB	Ward Class	Male -Tamil		13 Phase 2 students 9 female and 4 male

When we were recording these observations, we used Spradley's (1984) nine events, Space, Actor, Activity, Object, Act, Event, Time, Goal, and Feeling to make comprehensive notes in my field journal. We have rearranged those events into Spradley's earlier (1980) three "events"(Place, Actor and Activities) for my convenience during the analysis as follows: since our study focused on Sri Lankan culture, in which it emerged power differences are very important, we have added the "Power" as the fourth event

1. Place: Space, object
2. Actor:
3. Activities: Act, Event, Time, Goal, Feeling
4. Power

Findings

Teachers' statements consistently seemed to denote an assumption that the teachers have hierarchical power during the teaching-learning process. Respectful behaviours were most often witnessed as from the student towards the teacher rather than vice versa.

The teacher said "wait wait" at the time I observed that the student's face was not happy and was nervous, (Obs3). ..The teacher communicated to the students by using the word "hello" not by their name (Obs3)

Student behaviours also confirmed the power domains in between the teacher and students

There was a space between teachers and students while students were standing very close together. (Obs5)

When they saw the teacher all of them stood up and greeted the teacher (Obs3).

Several occasions we noticed a teacher trying to avoid the gap between the teacher and student. *Although they were seated very closely in a circle I felt there is a distance between teacher and students(Obs6)*

The teacher told the students to come very close. Students moved and made the circle smaller than earlier (Obs6).

Also, teachers changed their physical position in relation to the students, perhaps to reduce the perceived power differences in order to encourage students to talk

The teacher sat down with students and discussed (Obs2).
The teacher also was standing along with the students (Obs5).

Some students did not respond even though they knew the answer which we observed after the classes when they were sharing the answers within their peers. It may be the students were frightened in case their answer was wrong.

Students were timid. Most of the time students answered only when questions were asked.
Suddenly the teacher pointed a student(male) who was standing in the corner and asked a question. That student answered. After that teacher asked "why". When asking questions the student was looking at the roof or desk (no eye contact) (Obs4).

During Obs 6, we noted how students kept silent whilst a teacher scolded them stating that they were not prepared for the session. Their response seemed to be one of shame.

the teacher asked any question?? Nobody's talking. Then the teacher asked " any references" no one answers Then the teacher said, "I think you all here without reading the subject, if you come like this next time I will send you all from the class". All the students hung their heads without telling anything. Then the teacher listed out some names of the books (Obs6).

Students laughed in response to teachers' jokes but we did not once witness a student themselves cracking jokes in any of the eleven observations.

Sometimes, after the teacher said something, students were laughing (maybe joking) (Obs4).

Conclusion

Observations showed some evidence of formative mechanisms in use to identify gaps in learning but mostly related to achieving the correct answer with less evidence of identifying gaps in understanding. Although the activities of teachers and students expressed that the teachers have hierarchical power during the teaching-learning process, on several occasions teachers tried to reduce the gap between themselves and students by changing their physical position in relation to the students to encourage the students to talk.

Some students did not respond even though they knew the answer. Students seemed concerned, even sometimes frightened, that their answer was wrong. Humour seemed to be in one direction only, with only teachers free to make jokes, e.g. The relationships seemed to be dominated by cultural norms where respect was conferred upon the teacher and attempts to alter this were not successful as it affected the formative feedback process.

This study identified under-problematization of feedback practices, unaddressed cultural and historical complexities, alongside interest and willingness to develop post-colonial challenges to overly simplistic hegemonic "west is best" assumptions. Sri Lanka can be benefitted from WEIRD evidence-based theories such as dialogic feedback and learner transitions best when informed by and adapted to local cultural practices and knowledge during the feedback process.

References

1. Ajjawi R, Regehr G. When I say ... feedback. Med Educ. 2019;53(7):652-654. https://doi.org/10.1111/medu.13746
2. Bhattarai, M. (2007). ABCDEFG IS - the principle of constructive feedback. JNMA; journal of the Nepal Medical Association, 46(167), 151.
3. Dahlin, B., & Watkins, D. (2000). The role of repetition in the processes of memorising and understanding: A comparison of the views of German and Chinese secondary school students in Hong Kong. British Journal of Educational Psychology, 70(1), 65-84.
4. Dweck, C. S. (2012). Mindset. London: London : Robinson.
5. Guskey, T. (2005). Formative Classroom Assessment and Benjamin S. Bloom: Theory, Research and Implications. . Paper presented at the Annual Meeting of the American Educational Research Association Montreal, Canada. .
6. Guskey, T. R. (2007). Closing Achievement Gaps: Revisiting Benjamin S. Bloom's "Learning for Mastery". Journal of Advanced Academics, 19(1), 8-31. doi:10.4219/jaa-2007-704
7. Guskey, T. R. (2010). Formative Assessment: The Contributions of Benjamin S. Bloom. In H. L. Andrade & G. J. Cizek (Eds.), Handbook of formative assessment. New York, NY: Taylor & Francis.
8. Marambe, K. N., Vermunt, J. D., & Boshuizen, H. P. A. (2012). A Cross-Cultural Comparison of Student Learning Patterns in Higher Education. Higher Education: The International Journal of Higher Education and Educational Planning, 64(3), 299-316. Retrieved from http://libproxy.dundee.ac.uk/login?url=http://search.ebscohost.com/login.aspx?direct=true&db=eric&AN=EJ972403&site=ehost-live&scope=site. http://dx.doi.org/10.1007/s10734-011-9494-z
9. McCambridge J, Witton J, Elbourne DR. Systematic review of the Hawthorne effect: new concepts are needed to study research participation effects. J Clin Epidemiol. 2014 Mar;67(3):267-77. doi: 10.1016/j.jclinepi.2013.08.015. Epub 2013 Nov 22. PMID: 24275499; PMCID: PMC3969247.
10. Nicol, D. & Macfarlane-Dick. (2006). Formative assessment and self-regulated learning: a model and seven principles of good feedback practice. Studies in Higher Education, 31(2), 199-218.
11. Nicol, D. (2010). From monologue to dialogue: improving written feedback processes in mass higher education. Assessment & Evaluation in Higher Education, 35(5), 501-517.
12. 12. Patton, M. Q. (1999). Enhancing the quality and credibility of qualitative analysis. Health Serv Res, 34(5 Pt 2), 1189-1208
13. Scager, K., Akkerman, S. F., Pilot, A., & Wubbels, T. (2017). Teacher dilemmas in challenging students in higher education. Teaching in Higher Education, 22(3), 318-335. doi:10.1080/13562517.2016.1248392

14. Trompenaars, F., Hampden-Turner, C. . (2004). Managing People Across Cultures: Capstone Publishing Ltd.

15. Van Der Leeuw, R. M., Teunissen, P. W., & van Der Vleuten, C. P. M. (2018). Broadening the Scope of Feedback to Promote Its Relevance to Workplace Learning. Academic medicine : journal of the Association of American Medical Colleges, 93(4), 556. doi:10.1097/ACM.0000000000001962

16. Wiliam, D. (2011). What is assessment for learning? Studies in Educational Evaluation, 37(1), 3-14. doi:https://doi.org/10.1016/j.stueduc.2011.03.001

17. William, D. (2011). What is assessment for learning? Studies in Educational Evaluation, 37(1), 3-14. doi:10.1016/j.stueduc.2011.03.001

18. Williams, J. H., & Des Marais, E. A. (2016). Global Health Education. Soc Work Res, 40(1), 3-6. doi:10.1093/swr/svw001

19. Williamson, J. (2012). Teaching and learning in out-patient clinics. Clin Teach, 9(5), 304-307. doi:10.1111/j.1743-498X.2012.00545.

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Featured articles

The effect of Sodium-glucose cotransporter-2 inhibitors in the disease outcome among heart failure patients

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Heart failure (HF) is one of a leading health concerns with increasing prevalence. HF patients are classified according to their symptoms and the ejection fraction (EF). The classification includes;

- Reduced EF (EF<40%; HFrEF)
- Midrange EF (EF between 40% and 49%)
- Preserved EF (EF>50%; HFpEF)

The underlying pathophysiology of exacerbations among patients with HF is based on imbalance in sodium and fluid homeostasis. Conservation of sodium occurs among these patients even during appropriate volume or volume overload. This can lead to frequent hospital admissions. Furosemide and other loop diuretics are the mainstay of therapy. But the treatment with loop diuretics is challenged by its effect on neurohormonal activation. These mechanisms involve in preserving the intravascular volume by increasing renal tubular sodium reabsorption. This may lead to worsening of HF and mortality.

Sodium-glucose cotransporter-2 inhibitors (SGLT-2is) are oral hypoglycaemic agents which will benefit HF patients by reducing hospitalization and improving the outcomes among patients with reduced EF. The DAPA-HF trial involved treatment with dapagliflozin, and the results showed reduced hospitalization, reduced all-cause mortality, need for intravenous drugs, alleviated symptoms, and improved quality of life. These benefits were noticed among patients with diabetes and non-diabetic. A similar finding was noted in the EMPEROR-Reduced trial with empagliflozin treatment among HF patients. Based on this evidence and more extensive clinical trials, the European Society of Cardiology (ESC) has added dapagliflozin or empagliflozin with other standard treatment for HFrEF irrespective of diabetes in the management guidelines for HF

The following mechanisms showed the therapeutic benefit of empagliflozin in the management of HF;

1) A sustained natriuretic property of empagliflozin not depending on the degree of renal function and a synergistic effect with loop diuretics.
2) No worsening of potassium loss and improved handling of magnesium and uric acid.
3) A direct natriuretic effect and no osmotic diuresis.
4) The intravascular volume depletion not leading to significant RAAS or Sympathetic Nervous System activation or hypotension.

Loop diuretics have action at proximal tubular location compared to the movement of the SGLT2i's. This difference leads to increased sodium chloride delivery to the macula densa, and a small or absent neurohormonal response is initiated in SGLT2is therapy. This neurohormonal/renal/electrolyte safety profile of SGLT2i makes them a better adjuvant to loop diuretics rather than the thiazide diuretics. The required dose of loop diuretic can be reduced by adding the SGLT2 inhibitors.

SGLT2 inhibitors are contraindicated in;

- o History of allergic reaction to this group of drugs.
- o Pregnancy and breastfeeding.
- o eGFR <20 mL/min/1.73 m2.
- o Hypotension or Systolic Blood Pressure <95 mmHg.

This group of drugs is well tolerated, and the side effect profile is comparatively less. An increased risk of genital infection was noticed, and proper genital hygiene can prevent this. A baseline and on and off monitoring of blood sugar level and renal function test is recommended with SGLT2 inhibitors. Patients with established diabetes mellitus require monitoring for hypoglycaemia if they are on other anti-hyperglycaemic agents.

Heart failure patients encounter recurrent hospital admissions due to the exacerbations. This may occur due to the automatic activation of the neurohormonal system as a response to diuretic therapy. Adding SGLT2

inhibitors into the heart failure treatment regime has shown promising benefits in overcoming these challenges. A significant reduction in the disease exacerbation and hospital admissions were noticed after SGLT2 inhibitor treatment irrespective of the presence or absence of diabetes mellitus among patients.

References

1. Anker, S. D., Butler, J., Filippatos, G., Khan, M. S., Marx, N., Lam, C. S. P., Schnaidt, S., Ofstad, A. P., Brueckmann, M., Jamal, W., Bocchi, E. A., Ponikowski, P., Perrone, S. V., Januzzi, J. L., Verma, S., Böhm, M., Ferreira, J. P., Pocock, S. J., Zannad, F., … On behalf of the EMPEROR-Reduced Trial Committees and Investigators. (2021). Effect of Empagliflozin on Cardiovascular and Renal Outcomes in Patients With Heart Failure by Baseline Diabetes Status: Results From the EMPEROR-Reduced Trial. Circulation, 143(4), 337–349. https://doi.org/10.1161/CIRCULATIONAHA.120.051824

2. Brisco-Bacik, M. A., Ter Maaten, J. M., Houser, S. R., Vedage, N. A., Rao, V., Ahmad, T., Wilson, F. P., & Testani, J. M. (2018). Outcomes Associated With a Strategy of Adjuvant Metolazone or High-Dose Loop Diuretics in Acute Decompensated Heart Failure: A Propensity Analysis. Journal of the American Heart Association, 7(18), e009149. https://doi.org/10.1161/JAHA.118.009149

3. Butler, J., Handelsman, Y., Bakris, G., & Verma, S. (2020). Use of sodium-glucose co-transporter-2 inhibitors in patients with and without type 2 diabetes: Implications for incident and prevalent heart failure. European Journal of Heart Failure, 22(4), 604–617. https://doi.org/10.1002/ejhf.1708

4. Francis, G. S., Benedict, C., Johnstone, D. E., Kirlin, P. C., Nicklas, J., Liang, C. S., Kubo, S. H., Rudin-Toretsky, E., & Yusuf, S. (1990). Comparison of neuroendocrine activation in patients with left ventricular dysfunction with and without congestive heart failure. A substudy of the Studies of Left Ventricular Dysfunction (SOLVD). Circulation, 82(5), 1724–1729. https://doi.org/10.1161/01.cir.82.5.1724

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Case Reports

Acardius Acephalus - monochorionic twin pregnancy. A rare case presentation.

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Introduction

Acardiac twin is an extremely rare complication of monochorionic twin pregnancies with a known incidence of 1 % of monozygotic twins and 1 in 35 000 deliveries. It is due to the consequence of Twin Reversed Arterial Perfusion (TRAP). There is a lack of well-formed heart in one twin (Acardiac twin) and it gets perfusion from the donor twin (pump twin) through artery to artery and venous to venous placental anastomosis. Pump twin usually develops normally. Acardiac twin is always non-compatible with life. Pump twin usually develops normally but has a 9 % chance of getting karyotype abnormalities. Acardiac twin is always non-compatible for life. Acardiac twins have four morphological subtypes. Earlier this condition was known as chorioangiopagus parasiticus. (1),(2)

Acardius acephalus, is the most common type (60-75%).there is an absence of the head and upper torso and limbs in the acardiac twin with preservation of the lower limbs, genitalia and abdominal viscera

Acardius anceps is the most developed type,(20%) where rudimentary cranial structures present with the otherwise persistent trunk, limbs, and organs. But however, lacks even a rudimentary heart.

Acardius amorphus is the least differentiated type(5%) and comprises of an amorphous mass of bone, muscle, fat, and connective tissue. If rudimentary nerve tissue is present, it is then called Acardius myelantencephalus.

Acardius acormus the only developed structure is the fetal head, all other structures are essentially absent. Here the umbilical cord insertion is directly into the fetal head. Pathologically, rudiments of thoracic structures may be present. It is quite rare (10%).(3)(4)

Case report

A 23-year-old mother in her second pregnancy visited first time to the radiology department at 29 weeks of gestation for growth assessment. USG demonstrated monochorionic diamniotic twin pregnancy was detected with an anencephalic Pump twin which demonstrated cardiomegaly and bilateral pleural effusions suggestive of high output cardiac failure. (Figure 1)

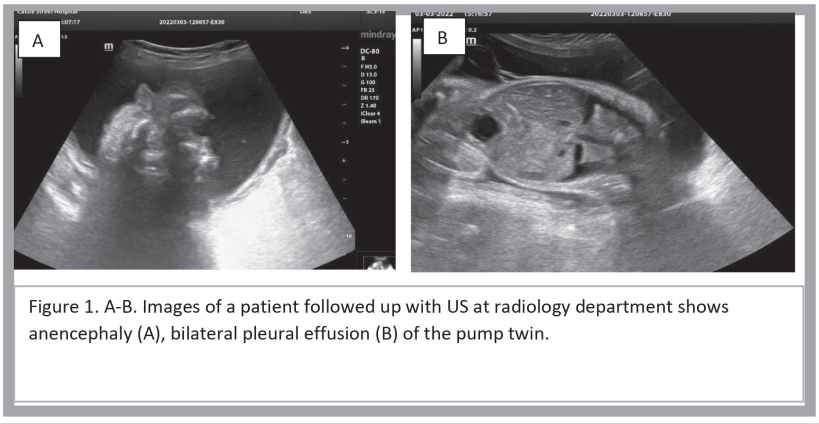

Figure 1. A-B. Images of a patient followed up with US at radiology department shows anencephaly (A), bilateral pleural effusion (B) of the pump twin.

Acardiac twin was identified which has only the lower segment including lower limbs, pelvis, and genitalia. There was significant edema with internal cystic areas suggestive of hypoperfusion. (Figure 2).The umbilical arteries of the Pump twin demonstrated normal flow. (Figure 3). However, no flow in the umbilical vessels of the Acardiac twin and no arterial anastomosis between the Pump twin was demonstrated.

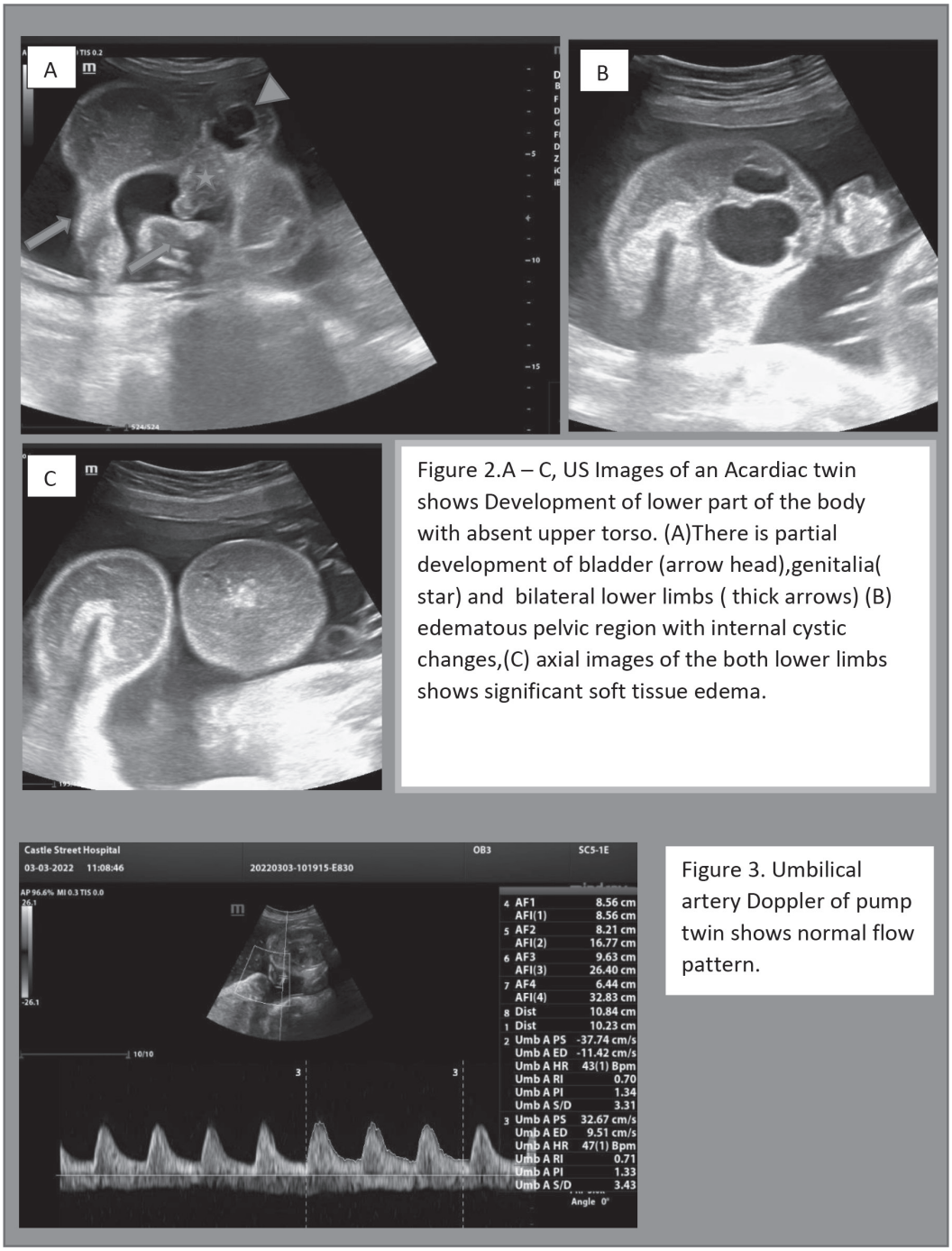

Figure 2.A – C, US Images of an Acardiac twin shows Development of lower part of the body with absent upper torso. (A)There is partial development of bladder (arrow head),genitalia(star) and bilateral lower limbs (thick arrows) (B) edematous pelvic region with internal cystic changes,(C) axial images of the both lower limbs shows significant soft tissue edema.

Figure 3. Umbilical artery Doppler of pump twin shows normal flow pattern.

Her clinical records state that her initial ward anomaly scan at 20 weeks scan pregnancy was misinterpreted her condition as single with an anencephalic fetus and meningomyelocele. It assumed that the partially developed cystic appearance of the Acardiac twin at the lower back of the pump twin must have been misinterpreted as meningomyelocele.

The pregnancy was terminated after the MDM decision at 30 weeks of gestation.

The scan finding was confirmed at the delivery of the anencephalic pump twin and Acardiac twin with the absence of development of the head and upper torso. The right hand was partially developed. (Figure 4)

Grossly, the placenta was single and diamniotic. The first twin's cord was of normal length, with a central insertion. The dysmorphic twin had a short, thin cord with a membranous insertion

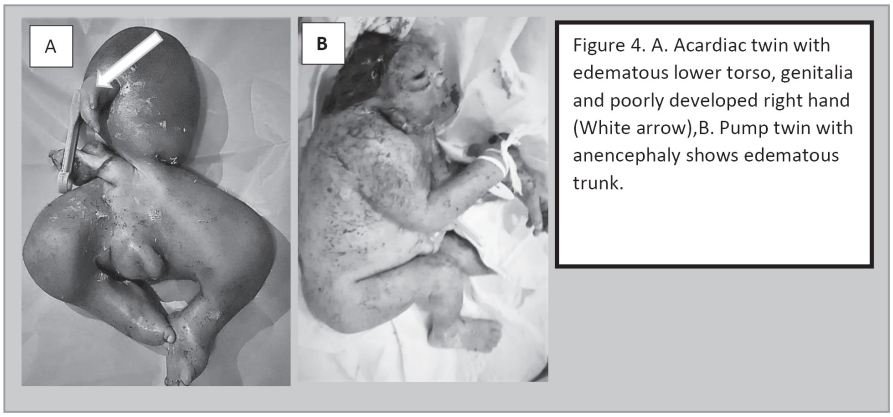

Figure 4. A. Acardiac twin with edematous lower torso, genitalia and poorly developed right hand (White arrow),B. Pump twin with anencephaly shows edematous trunk.

Discussion

Acardiac twinning is a very rare problem, One twin is usually structurally completely normal. The other is an abnormal mass of tissue, consisting usually of legs and a lower body, but no upper body, head, or heart. Because of the absent heart, the term "acardiac twin" has been used to describe this mass. The normal fetus is referred to as the "pump twin" because its heart is used to pump blood to the abnormal mass. The "acardiac twin" has no chance of survival.

Due to the absence of a beating heart, the acardiac does not send blood to any portion of the placenta, and all of its blood supply comes from and goes back to the circulation of the pump twin through unique vascular connections on the surface of the shared placenta. Arteries usually carry blood away from the fetus and toward the placenta to receive oxygen from the mother's circulation. When there is an "acardiac twin", the unique vascular connections allow blood in the artery to flow in the reversed direction (toward the acardiac fetus rather than away from it). Thus, the phrase "twin reversed arterial perfusion" (TRAP) sequence has been used to describe this condition. (5)

The normal "pump" twin faces the excess burden of having to send and receive blood to the acardiac mass as well as to its growing tissues. As such, the normal twin's heart has to work extra hard and is under a lot of stress. This can result in heart failure for the normal twin. The perinatal mortality is about 50–70% without intervention. These otherwise normal twins may die in utero (stillbirth) or die shortly after birth.(6)

A prompt initial diagnosis of monochorionic amniotic twin pregnancy and its complications are important. Therapeutic options are now available to secure otherwise normal pump twins in an Acardiac twin pregnancy. The treatment of acardiac anomaly presently relied on maximizing the term delivery's chance, prevention, CHF treatment in the healthy pump twin and interruption of the twin vascularization. Currently, management options included observation with close antepartum surveillance (conservative treatment) and surgical interventions. Intrafetal ablation is the treatment of choice for acardiac twins.(6)

References

1. Buyukkaya A, Tekbas G, Buyukkaya R. Twin reversed arterial perfusion (TRAP) sequence; Characteristic gray-scale and doppler ultrasonography findings. Iran J Radiol. 2015;12(3):1–3.

2. Masroor I, Jeelani S, Aziz A, Idrees R. Twin Reversed Arterial Perfusion Sequence: Assessing the Role of the Correct Imaging Modality in a Rare Clinical Entity. Cureus. 2018;10(7).

3. Rodeck CH WM. Fetal medicine, basic science and clinical practice. 2nd editio. Churchill Livingstone; 2008.

4. Vitucci A, Fichera A, Fratelli N, Sartori E, Prefumo F. Twin reversed arterial perfusion sequence: current treatment options. Int J Womens Health. 2020;12:435–43.

5. Hospitals UBC. TRAP Sequence or Acardiac Twin [Internet]. Available from: https://fetus.ucsf.edu/trap-sequence/

6. Nanthakomon T, Chanthasenanont A, Somprasit C, Manusook S, Pongrojpaw D, Suwannarurk K. Twin reversed arterial perfusion (TRAP) sequence: A case report and review of treatment. J Med Assoc Thail. 2015;98:S132–40.

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Case Reports

Accidental Hanging by Safety Rope- An Unusual Case

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Abstract:

An accidental hanging by ligature is reported. A 46-year-old man was found fully suspended by a rope tied to the chimney on the rooftop of his residence when he was trying to clean out the gutters with a leaf blower. Autopsy findings revealed a ligature mark on the surface of the neck, left anterior chest, and right anterior abdomen. Abrasions due to ligature are seen on the anterior neck and submental area. Internally bilateral horns of the thyroid cartilage and the left hyoid bone are fractured. No soft tissue hemorrhage is associated with these fractures. Lungs are congested. He did not possess any significant medical history and did not have any previous suicidal ideation or attempt or mental disorder. He was not under the influence of alcohol or any drugs. It was concluded that the man had died from asphyxia due to ligature hanging which occurred when he fell from a roof and became entangled in his safety rope. The manner of death is a classified accident.

Key Words: Accidental hanging, Asphyxia, Manner of death, Autopsy, Safety rope

Introduction

Asphyxial death is a common incident in forensic practice, and determining the manner of death is very important. The manners of death can be accidental, suicidal, homicidal, or natural due to the main methods of asphyxia. In such deaths, autopsy plays a significant role in solving the case; the scene investigation and collection of samples are also of importance[1, 2]

Among asphyxial deaths hanging is the leading cause of death1. Most hangings are suicidal [2 - 4.] Accidental hangings are uncommon and constitute nearly 5% of all hangings.[3, 5,] Deaths due to unintentional hanging among youth and adults are a well-known phenomenon, commonly occurring during autoerotic activity[6, 7, 8, 9.] Moreover, accidental hangings can be seen in rare cases, such as rounding the collar of a sweater10, hanging with a lanyard key chain[11,] hanging with a cloth belt[12,] and hanging with a hand strap of a garage door[13].

The present case is also a rare accidental hanging of an adult, which creates confusion about the manner of death.

Case report

A 46-year-old male was found hanging (fully suspended) by a rope tied to the chimney on the rooftop of his residence at around 16.15 pm. The decedent was last seen alive by his father around 1300 pm. At that time, the decedent was in good spirits and told his father that he would go up on the roof to clean the gutters with a leaf blower. His father left the residence to get groceries for dinner and returned around 1615 hours to find the decedent fully suspended in the backyard from a rope attached to the chimney on the roof. The rope was wrapped around the decedent's neck, and there appeared to be no signs of life (Fig 1). A rope was secured to the decedent's waist that had been strung through the belt loops of his pants. The front of his waist knotted the rope. The rope ligature then went upwards to his neck and was wrapped around only once with no knot around the neck (Fig 2). The rope looped over itself around the left side of his neck, and the neck exhibited abrasions on and around the ligature furrow. The rope continued to the top of the roof to the chimney, where it was knotted and secured around the chimney stack (Fig 3). A leaf blower was located on the rooftop. The decedent was clad in appropriately positioned clothing, and he was also wearing working gloves.

The decedent did not possess any significant medical history and had no previous suicidal ideation, previous attempts, or bouts with depression.

Autopsy Findings

The face is not congested, and the tongue is partially protruded. There are no petechial hemorrhages seen in the conjunctival palpebrae and oral mucosa. Received in place is a white woven rope measuring 1.5 centimeters in diameter. A portion of this rope is looped once around the neck, with one end of the loop cut and the other extending to a slipknot secured around the waist through the belt loops of the decedent's pants.

Underlying this rope is a ligature furrow around the neck and a faint furrow extending obliquely across the left chest and right abdomen. The deepest portion of the furrow around the neck is anteriorly located immediately superior to the thyroid prominence. On the lower border of the anterior furrow, a roughly 2 cm wide area of irregular abrasion is seen (Fig 4). The furrow extends approximately horizontally across the right lateral and posterior portions of the neck. The furrow on the left lateral neck extends superiorly to the point of suspension, about 3 cm inferior to the left ear. Growing inferiorly and to the right from the lower furrow is a faint depressed linear furrow extending across the left anterior chest and the right anterior abdomen, measuring up to 1.5 cm in width (Fig 5). In the right upper quadrant of the abdomen, just above the pant line, is a roughly 6*6 cm area of irregular blanching and erythema reflective of the adjacent slipknot in the rope as it passes through the belt loops (Fig 6). Associated with the ligature furrow around the neck are a 7 * 1.5 cm area of ecchymosis and faint abrasion on the submental surface extending from the midline to the right angle of the jaw (Fig 4).

Internally, bilateral horns of the thyroid cartilage and the left hyoid bone are fractured. No soft tissue hemorrhage is associated with these fractures.
Lungs are deep red and congested.
No other injuries are seen on the body.

Fig 1- The decedent was found fully suspended by a rope

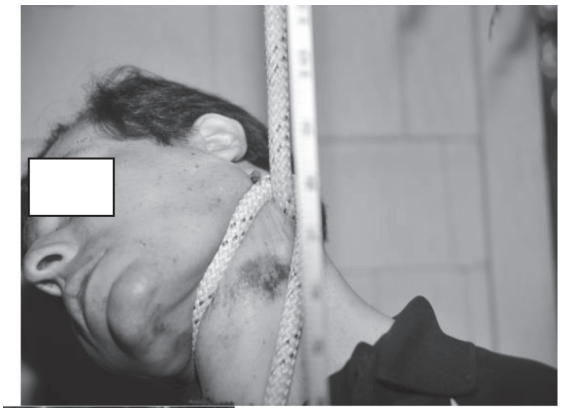

Fig 2 - rope was secured to the decedent's waist and wrapped around the neck with no knot.

Fig 3- The rope was knotted and secured around the rooftop's chimney stack and leaf blower.

Fig 4- Abrasion over the anterior neck and right submental area

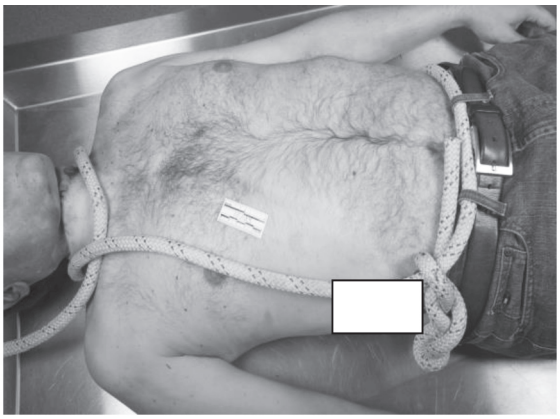

Fig 5- Faint depressed linear furrow extending across the left anterior chest and the right anterior abdomen

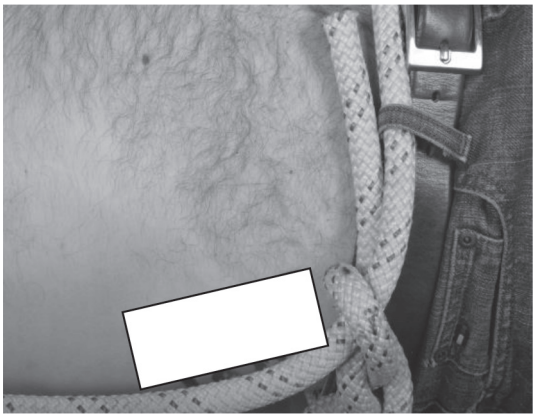

Fig 6- Erythema reflective of the adjacent slipknot in the rope

Discussion

Accidental hangings commonly fall into two groups: Infants and children; hanging occurs during play because of their curiosity to explore potentially dangerous things; in adults, it is associated with sexually deviant practices 2, 6-9. Many reported cases have involved accidents in infants and children 14-16 and autoerotic experiments 6-9. In addition, accidental hangings are usually associated with alcoholism10 or possible mental disturbance 17, for instance, elderly patients with dementia or Alzheimer's disease residing in a home for the elderly 18. Recently, accidental hangings of adults by lap-shoulder seat belts in motor vehicle accidents have also been reported19.In our case, it is different from the circumstances mentioned above. Here decedent is a young adult with a sound mind and not under the influence of alcohol or any other drugs.

A complete investigation of the scene of the incident and examination of the body in unnatural cases is critical. In Our case, no other injuries except those caused by the ligature. The death scene showed no signs of struggling or sexual behavior, according to the investigator involved in this case. The decedent had no history of mental illness, paraphilia, or sexual deviation. At the time of death, he was not intoxicated with ethanol. The decedent was wearing fairly good clothes and was also wearing working gloves. Further, the leaf blower was found on the top of the roof. There was no intention of suicide, and he did not leave any suicide notes. There is a remote possibility that others hung him, but an adult man without incapacitation by alcohol, any drugs, or injuries is very unlikely to hang by others. There was a reported case where two women hung an adult man while he was intoxicated with alcohol 19.
Our case decedent took precautions by securing himself with a rope, but unfortunately, it became ill-fated for him.

We concluded that the death had resulted from asphyxia due to ligature hanging which occurred when the decedent fell from a roof and became entangled in his safety rope. The manner of death is a classified accident.

Reference:

1) Derya Azmak. Asphyxial Deaths, A Retrospective Study and Review of the Literature. Am J Forensic Med Pathol 2006;27:134-144

2) DiMaio DJ, Dimaio VJM. Asphyxia. In: Forensic Pathology. Boca Raton, FL: CRC Press; 1993: 207-251

3) Cook CT, Cadden GA, Margolius KA. Death by Hanging in Western Australia. Pathology. 1995;27:268-272

4) Guarner J, Hanzlick R. Suicide by hanging, a review of 56 cases. Am J Forensic Med Pathol. 1987;8:23-26

5) Bowen DAL. Hanging: a review. Forensic Sci Int. 1982;20:247-249

6) Tournel G, Hubert N, Rouge C, et al. Complete autoerotic asphyxiation: suicide or accident? Am J Forensic Med Pathol. 2001;22: 180-183

7) Gosink PD, Jumbelic IM. Autoerotic asphyxiation in a female. Am J Forensic Med Pathol. 2000;21: 114-118

8) Uva Jl. Review: Autoerotic asphyxiation in the United States. J Forensic Sci. 1995;40: 574-581

9) Breitmeier D, Mansouri F, Albrecht K, et al. Accidental autoerotic deaths between 1978 and 1997: Institute of Legal Medicine, Medical School Hannover. Forensic Sci Int. 2003; 137: 41-44

10) Nurhantari Y, Asano M, Nushida H, et al. Accidental hanging by a sweater: An unusual case. Am J Forensic Med Pathol. 2002;23: 199-201

11) Denton JS. Fatal accidental hanging from a lanyard key chain in a ten-year-old boy. J Forensic Sci. 2002;47: 1345-1346

12) Hitosugi M, Takatsu A. An autopsy case of accidental hanging during home care. Nippon Hoigaku Zasshi. 2001;55:243-246

13) Schmidt U, Nadjem H, Pollak S. Death caused by accidental hanging in the hand strap loop of a garage door. Arch Kriminol. 2001;207:19-25

14) Amanuel B, Byard RW. Accidental asphyxia in bed in severely disabled children. J Paediatr Child Health 2000;36:66-68

15) Byard RW, Beal SM, Simpson A, et al. Accidental infant deaths and stroller-prams. Med J Aust 1996;165:140-141

16) Byard R, James R. Car window entrapment and accidental childhood asphyxia. J Paediatr Child Health 2001;37:201

17) Mason JK. Forensic medicine: an illustrated reference. London: Chapman and Hall, 1993:153-165

18) Osculati A, Fassina G. Two cases of accidental asphyxia by a neck compression between bed bars.Am J Forensic Med Pathol 2000;21:217-219

19) Wright RK, Davis J. Homicidal hanging masquerading as sexual asphyxia. J Forensic Sci 1976;21:387-389

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Case Reports

Button battery ingestion in a child: is it always necessary to perform urgent endoscopy?

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Abstract:

Button batteries are increasingly used in toys and electronic devices. They are considered highly hazardous since button batteries lodged in oesophagus can lead to oesophageal perforation, fistulation and in some children, fistulation into major thoracic blood vessels. The caustic injury to the oesophagus occurs rapidly within a duration as short as two hours. We report the case of a seven-year-old boy who presented following a potential button ingestion. The button battery was visualised within the pylorus of the stomach. The boy was managed with conservative approach since he remained asymptomatic and button battery had moved passed the oesophagus. This case highlights the importance of early evaluation of any child with button battery ingestion. Button battery ingestion must be recognised and treated promptly using a multidisciplinary approach to optimise outcomes for patients.

Introduction

Button batteries are highly hazardous and absence of physical symptoms does not rule out the risk of life-threatening complications. The unique size, shape and chemical properties of button batteries result in significant injury . When lodged in oesophagus of an otherwise asymptomatic child, a button battery can erode in to oesophagus and trachea, and lead to major haemorrhage following thoracic aortic rupture and rapid death.

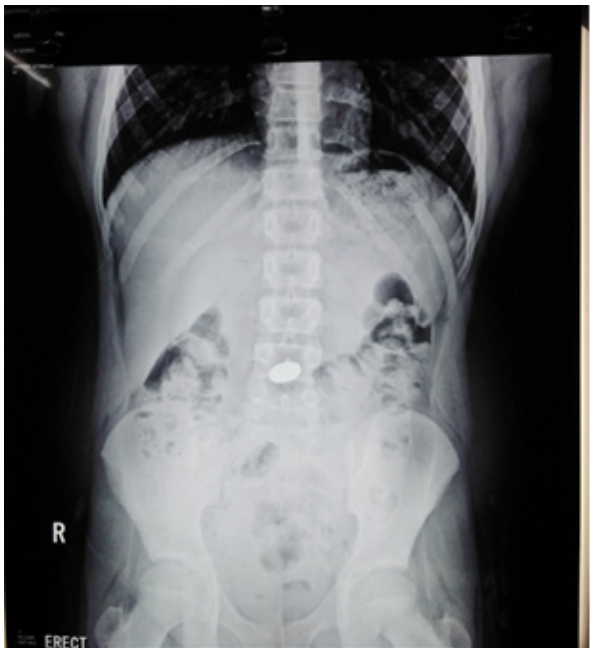
The chemicals present in button batteries include lithium, alkaline, zinc air, and silver oxide and they can lead to caustic injury. The common devices in which button batteries are found include watches, electronic toys, calculators, electronic locks and keys and remote controls . Studies have shown that even a used battery with a residual charge of at least 1.2 V has the ability to cause significant caustic injury1. In this report, authors have presented a seven-year-old boy who presented following ingestion of a button battery at home premises and subsequently, managed with conservative approach following timely and careful evaluation.

Case report

A seven-year-old child was brought three hours following a witnessed button battery ingestion by worried parents. The button battery from a toy car was given to the index child by the nine-year old elder brother and the history was not clear as if the child had chewed the battery before ingestion. The child had no past history accidental ingestions and had no reported behavioural difficulties. He did not have vomiting, drooling, epigastric or chest discomfort. He was able to cough and swallow without difficulty.

The general physical examination and systemic examination was normal. Urgent chest and abdominal radiography were performed and the button battery was visualised within the pylorus of the stomach (Figure 1). Upper gastrointestinal endoscopy was not performed provided that button battery was visualised distal to the oesophagus and the child remained asymptomatic. The child was reviewed in one week with repeat radiography to ensure that child did not have any evidence of gastrointestinal bleeding and buttery had passed down further beyond the stomach. Button battery was noted in faeces 8 days after the day of ingestion.

Figure 1: Button battery is shown within the proximal duodenum in the abdominal radiograph



Discussion

It is important to perform a thorough and timely evaluation on any child presenting with potential or witnessed button battery ingestion. Important history to note includes the time of ingestion, battery type and charge status, magnet co-ingestion, chemical composition, the number of batteries ingested and patient's comorbidities related to oesophagus . Analysing the symptomatology is not very helpful in unwitnessed ingestions since most children may present with symptoms that mimic either viral respiratory or gastrointestinal illness . Therefore, the complications are more likely in those with unwitnessed ingestions and those who remain asymptomatic until a complication develops .

The clinical presentation of button battery ingestion is variable. The common presenting symptoms include drooling, dysphagia and cough . However, absence of these symptoms neither rules out the likelihood of ingestion nor the risk of development of severe complications . Rarely, button battery can be aspirated in to the endobronchial tree and the child may present with acute onset severe respiratory distress .

Radiographic imaging is essential to make the diagnosis and the imaging should be performed without delay to reduce morbidity and mortality in all children less than 12 years with suspected or witnessed button battery ingestion . Clinical history and radiological diagnosis yield a sensitivity of almost 100% in confirming the final diagnosis and this highlights the importance of accurate risk assessment and radiography in these children.

Button batteries are highly hazardous and the reported complications include tracheoesophageal fistula, oesophageal stenosis, mediastinitis, vocal cord paralysis, pneumothorax, intestinal perforation and even, death following fatal haemorrhage and erosion in to major thoracic arteries .

Oesophageal button battery is an acute surgical emergency. Immediate endoscopic removal is indicated even if the child remains completely asymptomatic in the presence of an oesophageal button battery. Conservative approach is recommended to those in whom the possibility of oesophageal button battery is rule out and the course is asymptomatic. Repeat imaging is indicated in children who fail to demonstrate the battery by stool inspection 10-14 days after the day of ingestion3. Therefore, it is important that all asymptomatic children are followed up until the button battery is expelled from the gastrointestinal tract.

Conclusion

Urgent chest and abdominal radiography should be performed in any child younger than 12 years with suspected button battery ingestion. Urgent endoscopic removal should be performed in all asymptomatic children with batteries > 12 mm lodged in oesophagus. Batteries which have passed down beyond the oesophagus in otherwise asymptomatic children can be managed conservatively without needing urgent endoscopy. Timely and accurate risk assessment was crucial in optimising this child's clinical outcomes.

References

1. Jatana KR, Litovitz T, Reilly JS, Koltai PJ, Rider G, Jacobs IN. Pediatric button battery injuries: 2013 task force update. Int J Pediatr Otorhinolaryngol. 2013;77(9):1392-1399. 10.1016/j.ijporl.2013.06.006.

2. Litovitz T, Whitaker N, Clark L. Preventing battery ingestions: an analysis of 8648 cases. Pediatrics. 2010;125(6):1178-1183. 10.1542/peds.2009-3038.

3. Sethia R, Gibbs H, Jacobs IN, Reilly JS, Rhoades K, Jatana KR. Current management of button battery injuries. Laryngoscope Investig

Otolaryngol. 2021 Apr 15;6(3):549-563. doi: 10.1002/lio2.535. PMID: 34195377; PMCID: PMC8223456.

4. Buttazzoni E, Gregori D, Paoli B, et al. Symptoms associated with button batteries injuries in children: an epidemiological review. Int J Pediatr Otorhinolaryngol. 2015;79(12):2200-2207. 10.1016/j.ijporl.2015.10.003.

5. Labadie M, O'Mahony E, Capaldo L, et al. Severity of button batteries ingestions: data from French Poison Control Centres between 1999 and 2015. Eur J Emerg Med. 2018;25(4):e1-e8. 10.1097/MEJ.0000000000000528.

6. Ettyreddy AR, Georg MW, Chi DH, Gaines BA, Simons JP. Button battery injuries in the pediatric aerodigestive tract. Ear Nose Throat J. 2015;19(12):486-493. 10.1177/014556131509401207.

7. National Capital Poison Center. Fatal Button Battery Ingestions. 2023. https://www.poison.org/battery/fatalcases. Accessed June 17, 2023.

8. McLarty JD, Krishnan M, Rowe MR. Disk battery aspiration in a young child: a scarcely reported phenomenon. Arch Otolaryngol Head Neck Surg. 2012;138:680-682. 10.1001/archoto.2012.1097.

9. National Capital Poison Center . National Capital Poison Center Button Battery Ingestion Triage and Treatment Guideline. 2023. https://www.poison.org/battery/guideline. Accessed June 17, 2023.

10. Torrecillas V, Meier JD. History and radiographic findings as predictors for esophageal coins versus button batteries. Int J Pediatr Otorhinolaryngol. 2020;137:110208. 10.1016/j.ijporl.2020.110208.

11. Krom H, Visser M, Hulst JM, et al. Serious complications after button battery ingestion in children. Eur J Pediatr. 2018;177:1063-1070. 10.1007/s00431-018-3154-6.

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Case Reports

Internal Electric Current Mark - A Unique Case of High-Voltage Accidental Electrocution. Is It Underreported?

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Abstract:

A 34-year-old male succumbed to death due to a high voltage overhead power line electrocution at work. The autopsy revealed multiple external electrical and thermal injuries, contusions, and clavicular fractures sustained when thrown away following electrocution. In addition to these, we also found internal current marks on the liver and gall bladder in addition to the above injuries, which have never been reported in scientific journals before.

Key-Words:
Electrocution, High voltage, Internal current mark. Amperes, Resistance.

Introduction

Death following electrocution occurs due to the overwhelming conduction of electrical current through the body, most commonly causing ventricular dysrhythmia. Electrocutions usually need detailed investigation to determine how and why individuals became electrically energized, as the circumstances are challenging to solve[1].

Although most are used with the terms volts, a volt is a measure of electromotive force in a system. The amount of current flow per unit of time, or amperes, is the most critical factor to consider in human electrocution. Ventricular fibrillation could occur with 0.100 amperes, and ventricular standstill will happen with 2.00 amperes [1][2].

Electrocution is a specific hazard to people working routinely involved or working very close to electrical sources. Studies have revealed shown without any surprise that the major proportion of electrocution fatalities occurs among electricians and electrical assistants[5][6][7] and among utility workers[6][8]. A reasonable number of construction and manufacturing industry employees also died annually due to electrocution[5][9][10][11].

Our case also highlights an incident where a young construction worker was electrocuted with a high voltage overhead power line while at work, but injuries give us a different perspective. We found internal current marks on the liver and gall bladder in addition to standard external injury patterns. In our opinion, this has never been reported in scientific journals before. Only one reported case by Anders S et al. where an electrician committed suicide by low voltage prolonged electrocution using a time switch after oral ingestion of diazepam. They found injury over the pleura parietalis and the inner surface of the thoracic cavity[12].

Case report

A 34-year-old male worked for a private contractor for ten years as a framer. The deceased guided a load placed on the ground by a top-loading crane. He held onto a chain when the crane arm swung into an overhead power line electrocuting the subject. The subject was thrown and initially alert. He became unconscious shortly after. The subject was immediately transported to a medical center, where he remained comatose. The subject was declared brain-dead after three days. Tissue procurement was performed.

Autopsy Findings

The body was a normally developed, well-nourished male adult, appearing at the stated age of 34 years,

weighing 188 pounds, and measuring 5 feet 8 inches. Severe rigor mortis was observed in the upper and lower extremities and the jaw. Livor mortis is purple and fixed on the dependent part of the back of the body.

External Injuries

1) Burn marks were noted over the mid-sternal and lower sternal areas of the anterior chest (Figure 1). Wounds appeared irregular, yellow, and firm with white-pink circumference and sparse charring of the central area.
2) A 7 x 8-inch area of punctuating, circular, and confluent burns are on the central back (Figure 2).
3) 11 burned lesions on the buttock, varying in size and shape with the white-pink periphery and light to dark brown central scabs (Figure 2).
4) Burned lesions on the fingers and palmar area of the right hand were also noted. (Figure 3).
5) contusions on the chest, right upper, and forearm.

Internal Injuries

1) There was a non-displaced right midclavicular fracture.
2) There were two circular black lesions with a white-pink circumferential border measuring 1 centimeter in diameter; one was on the inferior left lobe of the liver, and one was on the inferior quadrate lobe of the liver (Figure 4).
3) There was a 2-centimeter horizontal linear transmural burn on the inferior gall bladder (Figure 4).

The liver weighs 1920 grams and has a smooth glistening capsule. Cut surfaces were uniformly yellow-tanned and had the usual landmarks. Microscopically liver showed evidence of thermal injury and moderate to severe steatosis.

He had no significant pre-existing medical conditions, and his toxicology results were uneventful.

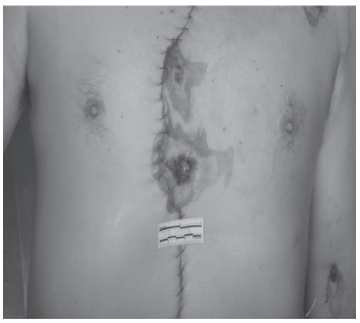

Fig 1 – Electric burns on the anterior chest.

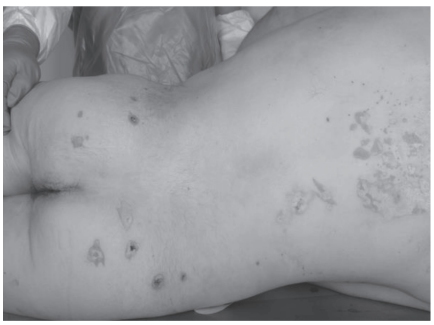

Fig 2- Electric burns on the back of the body.

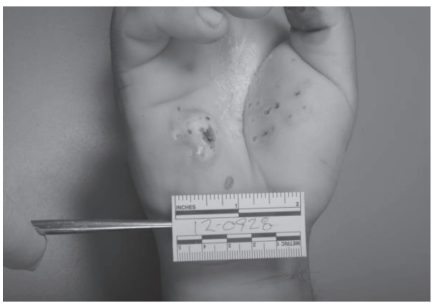

Fig 3- Electric burns on the right hand.

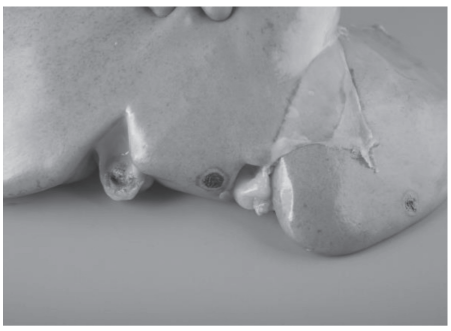

Fig 4- Burns over the liver and gall bladder.

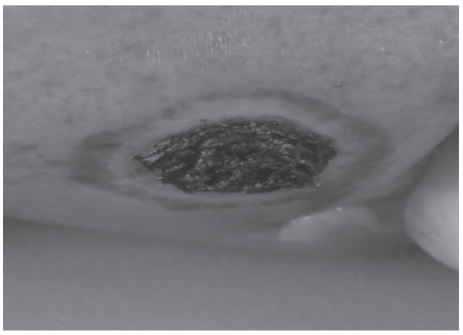

Fig 5- Closer view of the burn over the inferior left lobe of the liver.

Discussion

As per available statistical data, occupational fatalities secondary to coming into contact with electricity account for approximately 9% of all deaths in the construction industry and is the fourth leading cause of death in this industry [13].

Total amperage, or the amount of current flow, is the critical factor determining fatality due to

electrocution. It is directly related to the voltage and inversely associated with the resistance. “Voltage” is the measure of the electromotive force, and “Ohms” is the resistance to the conduction of electricity. This is scientifically expressed in the formula of [14] A = V/R

As one would expect, most of the fatalities due to electrocution happen due to high-voltage electrocution, and most of them result in electrical burns on the body. Still, electrical burns on the body are found in half or less than half the cases of low-voltage electrocution. In contrast to low-voltage burns, high-voltage burns can be extremely severe and result in the body’s charring. One would note numerous individual and confluent areas of third-degree burns during autopsy. The multiple minor burns are due to the arching of the current [14]. In high-voltage electrocution, internally, one might be able to see asphyxial findings, hemorrhagic pulmonary edema, right heart dilatation, and visceral petechiae[15]. Sometimes, internal organs may rupture [14] and tears in major arteries[15]. Except for one case by Anders S et al., there are no reported cases in the scientific literature about internal current marks. Anders S et al. describes a suicide of an electrician by prolonged low-voltage electrocution and the internal current marks on the pleura parietalis and the inner surface of the thoracic cavity[12].

Electrical burns frequently result in high-temperature burns, which produce characteristic findings of severe thermal denaturation of collagen. The epidermis is often elevated with micro blisters resulting from the cooking effect of the tissue by the high voltage of electricity and represents channels through which steam exited. Nuclei of epidermal cells at the site of an electrical burn frequently show stretching and narrowing of the contour to produce a palisade-type (Streaming of the nuclei) appearance[16].

The skin is the main barrier in terms of resistance to electrocution in humans. Human dry skin can have a resistance of 100,000 ohms and dry and calloused skin up to a million ohms[17]. Moist skin has a resistance of 1000 ohms or less[14]. Stanly Rush et al. studied the resistivity of body tissues at low frequencies in dogs, and their results are shown in Table 1[18].

Table- 1 Mean Resistivity (ohms-cm) of the dog tissues by Rush, Abildskov, and McFee

Tissues	Resistivity (ohms-cm)
Blood	162
Liver	700
Heart	563
Lungs	2100
Fat	2500
Skeletal muscle	2300
Trunk	465

C. Mirescu et al. studied electrophysiological evaluation of the normal and the intoxicated rat liver. They found that electrical resistance was higher in the intoxicated group. The leading cause for the increased electrical resistance of the intoxicated group could be liver steatosis due to intoxication with carbon tetrachloride (9). Fat generally has a lower electrical conductivity than substances containing water. Also, the collagen ridges along the portal structures are very poor in water content. The low perfusion due to steatosis is another cause of low electrical conductivity[19].

In the case we are discussing here, we found lesions on the liver and gall bladder, macroscopically and microscopically consistent with thermal injury. The liver shows moderate to severe hepatosteatosis. Even though livers have relatively low electrical resistance (700 ohms-cm), in the present case, due to steatosis liver, would have high resistance because fat tissues have high electrical resistance (2500 ohms-cm). We could explain that the internal electrical mark on the liver and gall bladder is because of the high resistance on the liver due to steatosis.

We concluded that the death had resulted from electrocution, and the manner of death was classified as an accident.

References

1] David Dolinak and Evan W.Matshes, Forensic Pathology: Principles and practice, 1st ed. Elsevier Academic press, Burlington, Ma, USA, 2005. 250-252 p.

[2] R. K. Wright and J. H. Davis, “The investigation of electrical deaths: a report of 220 fatalities.,” J Forensic Sci, vol. 25, no. 3, pp. 514–21, Jul. 1980.

[3] A. J. Taylor, G. McGwin, F. Valent, and L. W. Rue, "Fatal occupational electrocutions in the United States.," Inj Prev, vol. 8, no. 4, pp. 306–12, Dec. 2002, doi: 10.1136/ip.8.4.306.

[4] "Us Bureau of Labor Statistics. National census of fatal occupational injuries, 1998.," Washington, DC: US Department of Labor, 1999. Available at https://www. https://www.bls.gov/news.release/history/cfoi_08121999.txt. Accessed on12/10/2022

[5] A. J. Taylor, G. McGwin, G. G. Davis, R. M. Brissie, and L. W. Rue, "Occupational electrocutions in Jefferson County, Alabama.," Occup Med (Lond), vol. 52, no. 2, pp. 102–6, Mar. 2002, doi: 10.1093/occmed/52.2.102.

[6] "National Institute of Occupational Safety and Health. Worker death by electrocution, a summary of NIOSH surveillance and investigative findings. Report No 98- 131.," Cincinnati, Oh: NIOSH. Available at htttps://www. https://www.cdc.gov/niosh/docs/98-131/pdfs/98-131.pdf. 9Accessed on 13/11/2022)

[7] D. M. Fatovich, "Electrocution in Western Australia, 1976-1990.," Med J Aust, vol. 157, no. 11–12, pp. 762–4, doi: 10.5694/j.1326-5377.1992.tb141279.x.

[8] D. Loomis, V. Dufort, R. C. Kleckner, and D. A. Savitz, "Fatal occupational injuries among electric power company workers.," Am J Ind Med, vol. 35, no. 3, pp. 302–9, Mar. 1999, doi: 10.1002/(sici)1097-0274(199903)35:3<302::aid-ajim11>3.0.co;2-b.

[9] J. E. Jones, C. W. Armstrong, C. D. Woolard, and G. B. Miller, "Fatal occupational electrical injuries in Virginia.," J Occup Med, vol. 33, no. 1, pp. 57–63, Jan. 1991, doi: 10.1097/00043764-199101000-00015.

[10] T. Ore and V. Casini, "Electrical fatalities among U.S. construction workers.," J Occup Environ Med, vol. 38, no. 6, pp. 587–92, Jun. 1996, doi: 10.1097/00043764-199606000-00009.

[11] A. Suruda and L. Smith, "Work-related electrocutions involving portable power tools and appliances.," J Occup Med, vol. 34, no. 9, pp. 887–92, Sep. 1992.

[12] S. Anders, J. Matschke, and M. Tsokos, "Internal current mark in a case of suicide by electrocution.," Am J Forensic Med Pathol, vol. 22, no. 4, pp. 370–3, Dec. 2001, doi: 10.1097/00000433-200112000-00007.

[13] C. A. Janicak, "Occupational fatalities due to electrocutions in the construction industry.," J Safety Res, vol. 39, no. 6, pp. 617–21, 2008, doi: 10.1016/j.jsr.2008.10.007.

[14] V. J. M. D. M. Dominick J. Di Maio, Electrocution. In: Forensic Pathology.3rd Edition, CRC Press, Inc, 1993.

[15] G. G. Wright RK, Handbook of Forensic Pathology. Chapter: Electrical Injuries & Lightning. 1990.

[16] E. R. D. Barry D. Lifschultz, "Electrical and lightning injuries.," in Spitz and Fisher's Medicolegal Investigation of death, Charles C Thomas Publisher, Illinois, 1993, pp. 516–527.

[17] J. M. Bruner, "Hazards of electrical apparatus.," Anesthesiology, vol. 28, no. 2, pp. 396–425, doi: 10.1097/00000542-196703000-00023.

[18] S. RUSH, J. A. ABILDSKOV, and McFEER, "Resistivity of body tissues at low frequencies.," Circ Res, vol. 12, pp. 40–50, Jan. 1963, doi: 10.1161/01.res.12.1.40.

[19] C.-L. R. C. Mirescu, "Electrophysiological evaluation of the normal and intoxicated rat liver: Electrical resistance measurements," Annals of RSCB, Vol. XV, no. Issue 2, pp. 400–403, 2010.

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Case Reports

Correction of Transverse Mandibular Deficiency with a Hybrid Distractor- A Case Report

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Introduction

Transverse mandibular deficiency is one of the common skeletal problems associated with narrow basal and dentoalveolar bones. Transverse mandibular deficiency is marked by facial asymmetry, scissor-bite, a tapered mandibular arch, crowded misaligned anterior teeth, and a narrow inter-canine width. In patients with severe mandibular transverse discrepancies, mandibular midline distraction osteogenesis (MMDO) is a surgical procedure that widens the jaw by progressive traction and gradually separates the mandibular symphysis (1). Rosenthal first introduced MMDO in 1951, and Guerrero et al. made modifications to it in the 1990s (2). Different distraction appliances (bone-borne, tooth-borne, or hybrid distraction appliances) can be used to treat MMDO (1). Since the hybrid distraction device is anchored to both the bone and teeth, it combines the benefits of both tooth- and bone-borne devices.

Case history

A 24-years old female patient presented to the oral and maxillofacial surgical unit, Teaching hospital Karapitiya with a complaint of right side lower facial asymmetry and altered occlusion. The patient was born full term to non-sanguineous parents with no significant family or antenatal history. Clinical examination showed a right-side facial asymmetry due to mandibular deficiency on the affected side and a Class II, division 1 malocclusion associated with a scissor bite on the same side.

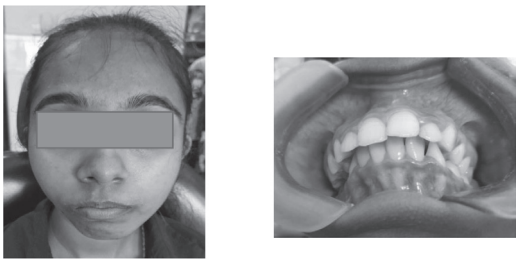

Figure 1. Pretreatment facial photographs and Pretreatment intraoral photographs.

Preoperative posteroanterior cephalometric measurements showed that the mandibular (biantegonial) and mandibular inter canine widths were less than the Ricketts adult norms. At the multidisplinary team meeting decided to widen the right side mandible with a hybrid distractor to address the patient's facial asymmetry and scissor bite.

Under general anesthesia, a mid-symphyseal osteotomy was carried out with taking care to protect the teeth. A bone-tooth-supported distractor was adjusted and evaluated for expansion right away after the osteotomy (Figure 5). The patient was given antibiotics and a 0.012% chlorhexidine rinse during a 4-day latency period. Following the latency period, the distraction procedure was initiated once daily at a pace of 1 mm. Mandibular width increased by around 7 mm. The distractor was withdrawn after the consolidation period while under general anesthesia.

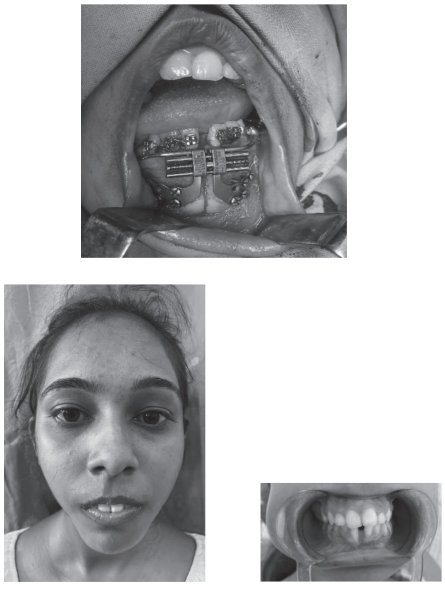

Figure 2.intraoperative photograph and Post treatment facial photographs.

Discussion

Symphyseal widening devices can be either intraoral or extra oral. Intraoral devices, which can be divided into three categories: tooth-supported, bone-supported, and hybrid, are frequently employed due to aesthetic desires. Hybrid expansion devices have a larger ability to achieve proportionate movement, which means more stable outcomes can be anticipated. Given that it is anchored to both the bone and the teeth, a hybrid distraction appliance combines the benefits of both bone-borne and tooth-borne appliances (3). The position of the distractor and the direction of the distraction vector are crucial factors because they might affect how the distraction gap looks. The distraction should result in a pure translation of the bone segments if the force is given close to the mandible's center of resistance, and the distraction gap should have parallel edges. The two bone segments may, however, rotate if the force is applied above the mandible's center of resistance. This will cause a disproportionally bigger gap in the alveolar region than in the basal region.

Straight and step lines are the two types of osteotomy for symphyseal widening. Guerrero et al. suggested using a parasymphyseal straight osteotomy on the ipsilateral side to treat unilateral symphyseal widening, however, medical experts have recommended using cross-arch elastics in combination with a straight mid-symphyseal osteotomy to stop bilateral expansion (2). The symphyseal region showed the largest widening when the effects of MSDO were evaluated anteroposteriorly, and the widening impact steadily diminished from anterior to posterior. This patient's mandibular intercanine width had increased from 23 mm at baseline to 30 mm just after distraction.

Wound dehiscence, pressure ulcers, distracting appliance-related issues, infection, and tooth damage are the most frequently reported side effects following MMDO with hybrid appliances (4). Although periodontal and dental morbidity following MMDO with a hybrid appliance appears to be temporary and restricted to the distraction and consolidation period (5), tooth mobility and expansion of the periodontal ligament next to the osteotomy have previously been documented after MMDO. During the distraction phase, MMDO produces rotational motions of the mandibular condyles. Temporomandibular joint pain following MMDO is infrequent, and in the included studies, the pain was relieved with physiotherapy and the removal of the hybrid appliance (6).

Conclusion

The safe method known as mandibular symphyseal distraction osteogenesis (MSDO) can be carried out with either local or general anesthesia. Patients with tooth-jaw discrepancies and transverse mandibular hypoplasia of more than 7 mm may be successfully treated with no evidence of relapse in the long-term follow-up.

References

1. Alkan A, Ozer M, Baş B, Bayram M, Celebi N, Inal S, Ozden B. Mandibular symphyseal distraction osteogenesis: review of three techniques. Int J Oral Maxillofac Surg. 2007 Feb;36(2):111-7.

2. Guerrero CA, Bell WH, Contasti GI, Rodriguez AM. Mandibular widening by intraoral distraction osteogenesis. Br J Oral Maxillofac Surg. 1997 Dec;35(6):383-92

3. Niculescu JA, King JW, Lindauer SJ. Skeletal and dental effects of tooth-borne versus hybrid devices for mandibular symphyseal distraction osteogenesis. Angle Orthod. 2014 Jan;84(1):68-75

4. de Gijt JP, Vervoorn K, Wolvius EB, Van der Wal KG, Koudstaal MJ. Mandibular midline distraction: a systematic review. J Craniomaxillofac Surg. 2012 Apr;40(3):248-60.

5. Kewitt GF, Van Sickels JE. Long-term effect of mandibular midline distraction osteogenesis on the status of the temporomandibular joint, teeth, periodontal structures, and neurosensory function. J Oral Maxillofac Surg. 1999 Dec;57(12):1419-25; discussion 1426.

6. Gunbay T, Akay MC, Aras A, Gomel M. Effects of transmandibular symphyseal distraction on teeth, bone, and temporomandibular joint. J Oral Maxillofac Surg. 2009 Oct;67(10):2254-65.

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Case Reports

Metatropic Dysplasia; A Case Report

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Abstract:

Metatropic dysplasia is a rare severe form of spondylo-metaphyseal dysplasia characterized by dumb bell like configuration of the long bones, short limbs, severe scoliosis, high dorsal kyphoscoliosis, joint hyper mobility, a narrow but normal length thorax and occasionally a coccygeal appendage similar to a tail. The authors report a seven-month-old male infant with physical phenotype and radiological features highly suggestive of metatropic dysplasia. Genetic studies could not be performed due to lack of financial resources.

Introduction

Metatropic dysplasia is a short limb skeletal dysplasia characterized by a dumb bell like configuration of the long bones, short limbs, severe scoliosis, high dorsal kyphoscoliosis, joint hyper mobility, a narrow but normal length thorax and occasionally a coccygeal appendage similar to a tail. It is a severe form of spondylo-metaphyseal dysplasia . It is a rare condition, with approximately 100 cases reported in the medical literature. There are several genetic variants including a non-lethal type with autosomal recessive transmission, a nonlethal dominant type and a lethal type with death before or shortly after birth. However, most cases occur sporadically and genotype-phenotype correlations are not clear1. The authors report an infant with physical and radiological findings are consistent with a diagnosis of severe spondylo-metaphyseal dysplasia (metatropic dysplasia).

Case report

A seven-month-old infant was admitted following a lower respiratory tract infection. He was the second born to healthy non consanguineous parents. Five- year- old elder sibling was healthy. Antenatal ultrasound scan showed multiple limb anomalies suggestive of a skeletal dysplasia. The birth was uneventful and birth weight was 2.54 kg.

The anthropometric parameters (weight, occipitofrontal circumference and height) at seven months remained less than -3SD for the chronological age. General examination revealed short limbs and hypoplasia of left hemiface. Fontanelles were widely open. Limbs were shortened. There were no fractures, increased fragility of bones, no club foot, cleft lip/palate or any other deformities. Fingers in hands could not be fully extended. Spine examination revealed kyposcoliosis with a caudal appendage (Figure 1). Cardio respiratory system examination findings were apart from a narrow bell-shaped chest.

Figure 1 - Kyphoscoliosis and caudal appendage

Radiological findings included hypertrophy of metaphyseal plates in all long bones with dumbbell shaped long bones, short, squared shaped iliac bones, with narrowed greater sciatic notch (Figure 2), odontoid hypoplasia (Figure 4), kyphoscoliosis in the spine, with platy spondyles, short ribs with flared, cupped anterior ends (Figure 3), and wide inter vertebral disk spaces. Phalanges were hourglass

shaped. Radiological diagnosis was compatible with severe spondylo-metaphyseal dysplasia. 2 D echocardiography was normal.

Serum Alkaline Phosphate, blood urea, serum calcium, serum electrolytes, liver functions and Full Blood Count were normal. USS abdomen and brain did not show any abnormalities.

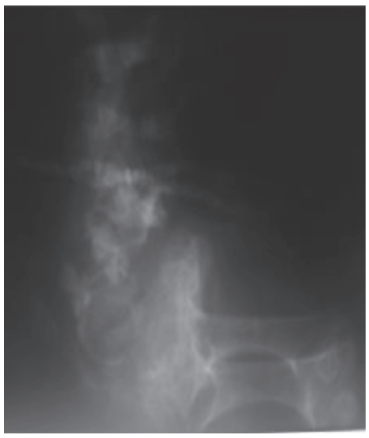

Figure 2 – Short, squared shaped iliac bones, with narrowed greater sciatic notch

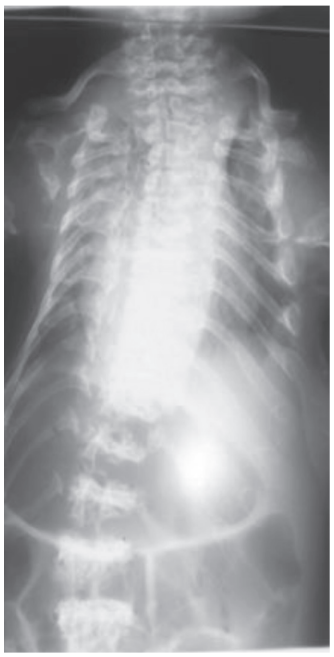

Figure 3 - Short ribs with flared, cupped anterior ends.

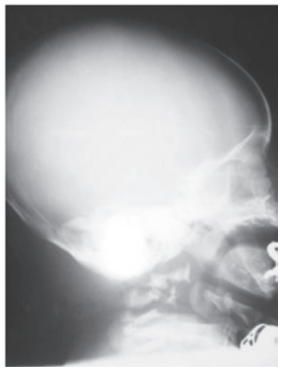

Figure 4 – Odontoid hypoplasia

Discussion

Metatropic dysplasia is a rare severe form of spondylo-metaphyseal dysplasia with progressive dwarfism and characteristic clinical and diagnostic radiographic findings. In children with metatropic dysplasia, the notable physical features include a long trunk with disproportionately short extremities (dumbbell-like configuration of the long bones) and a narrow chest of normal length. Often a 1-3 cm long tail-like appendage is visible at the level of coccyx . The joints are prominent with decreased mobility and fingers and toes are long. The face is uncharacteristic although a prominent forehead and flattened nasal bridge may be evident in infancy. The proportions change during childhood with relative shortening of the trunk due to progressive kyphosis and relatively long extremities . Chest deformity and flexion contractures are other associated findings. Hyper extensibility of the fingers may be present. Intellectual development is normal.

Variants in TRPV4 gene are known to cause severe phenotypes of metatropic dysplasia . Severe phenotypes are associated with severe skeletal abnormalities and neurological complications . The progression of kyphosis and other skeletal abnormalities differ among affected patients. Physiotherapy and orthopaedic interventions form part of multidisciplinary treatment for patients with metatropic dysplasia . Staged surgical corrections are often needed to improve quality of life . The reported complications include compression of the cervical cord, limitation of motor development and severe kyphoscoliosis10.

Conclusion

The report describes an infant with physical phenotype and radiological features that characterise metatropic dysplasia. It is an extremely rare severe form of spondylo-metaphyseal dysplasia. Genetic studies are indicated for identifying the underling genotype and confirming the genetic diagnosis.

References

1. Z Gucev, G Kalcev, N Laban, et al. Characteristic Diagnostic Clues of Metatropic Dysplasia: The Lumbothoracic Humpback with Dumbbell Appearance of the Long Bones. Balkan J Med Genet. 2018 Dec 31;21(2):35-38. doi: 10.2478/bjmg-2018-0025.

2. Song HR, Sinha S, Song SH, Suh SW. A case of metatropic dysplasia: operative treatment of severe kyphoscoliosis and limb deformities. Oman Med J. 2013 Nov;28(6):445-7. doi: 10.5001/omj.2013.123.

3. Bargiacchi S, Della Monica M, Biagiotti R, Andreucci E, Ciabattoni S, Poggi P. Metatropic dysplasia in third trimester of pregnancy and a novel causative variant in the TRPV4 gene. Eur J Med Genet. 2017;60(7):365–368.

4. Akhter S, Rahman S, Momen M, Karim M, Kawser C. Metatropic dysplasia in children. Mymensingh Med J. 2008 Jan;17(1):93-7.

5. Beck M, Roubicek M, Rogers JG, Naumoff P, Spranger J.: Heterogeneity of metatropic dysplasia. Europ. J. Pediat. 140: 231-237, 1983. PubMed ID : 6628444

6. Dai J, Kim OH, Cho TJ, et al. Novel and recurrent TRPV4 mutations and their association with distinct phenotypes within the TRPV4 dysplasia family. J Med Genet. 2010;47:704-709.

7. Schindler A, Sumner C, Hoover-Fong JE. TRPV4-Associated Disorders In: Pagon RA, Adam MP, Ardinger HH, et al., eds. GeneReviews(R). Seattle (WA): University of Washington, 1993:1993-2018.

8. Graversen L, Haagerup A, Andersen BN, Petersen KK, Gjørup V, Gudmundsdottir G, Vogel I, Gregersen PA. Novel TRPV4 variant causes a severe form of metatropic dysplasia. Clin Case Rep. 2018 Jul 20;6(9):1774-1778. doi: 10.1002/ccr3.1598.

9. Unger S, Lausch E, Stanzial F, et al. Fetal akinesia in metatropic dysplasia: the combined phenotype of chondrodysplasia and neuropathy? Am J Med Genet A. 2011;155A:2860-2864.

10. Kleszczyński J, Błoch M, Hofbauer K, Dominiak P. Metatropic dysplasia in four-year-old boy--physiotherapy and orthopaedic care problems--case report. Ortop Traumatol Rehabil. 2012 May-Jun;14(3):289-302. English, Polish. doi: 10.5604/15093492.1002258.

11. auer JM, Ditro CP, Mackenzie WG. The Management of Kyphosis in Metatropic Dysplasia. Spine Deform. 2019 May;7(3):494-500. doi: 10.1016/j.jspd.2018.09.007.

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Case Reports

A rare case of abrupt onset vascular lump in the newborn; neonatal kaposiform haemangioendothelioma

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Abstract:

Kaposiform hemangioendothelioma is a rare vascular tumor in children. It is known to be originated from the lymphatic endothelium of skin and infiltrated into subcutaneous tissues. Early diagnosis is important given the risk of Kasabach-Merritt syndrome. We report the case of a newborn who developed a rapid onset vascular lump within the first few hours of life. The diagnosis of neonatal onset kaposiform haemangioendothelioma was diagnosed based on clinical evolution of the lump and radiological findings.

Introduction

Kaposiform hemangioendothelioma (KHE) is a rare vascular tumor in children . About fifty percent of the cases present at birth and the rest will appear in early childhood. The prevalence of KHE is equal in both females and males with a slight male preponderance . It is found to be originated from the lymphatic endothelium of skin and infiltrated into subcutaneous tissue . KHE usually has a benign progression and distant metastasis is rare. The complications of Kaposiform hemangioendothelioma are due to local invasion, compressive effects, and the life-threatening consumptive coagulopathy known as Kasabach-Merritt syndrome , . We report a newborn who had an uncomplicated birth and was diagnosed to be having Kaposiform hemangioendothelioma within first few hours of life. The report highlights the importance of considering KHE as one of the differential diagnoses when a newborn presents with a rapidly enlarging vascular lump following a normal newborn examination at birth.

Case Report

The paediatrician was called to see an abrupt onset vascular lump over the left lateral thigh in a six-hour-old newborn. The baby was born in good condition and intramuscular vitamin K was given to the left lateral thigh. Newborn examination at birth was normal and careful examination did not reveal any vascular naevi anywhere in the body at the time of birth. Initially, the lump was felt to be a haematoma; however, vitamin K injection site was clearly seen adjacent to the vascular lump and there was no evidence of bleeding tendency at the injection site. Antenatal and family history were unremarkable. There was no family history of haematological or vascular diseases.

The lump was erythematous, irregular, and 3.8 cm x 3.6 cm in diameter (Figure 1). No other similar lumps or any other evidence of bleeding manifestations were seen. An urgent ultrasound scan of the lump revealed evidence of haemangioma with two well-developed feeding arteries. Based on the clinical evolution of the lump and radiological findings, kaposiform haemangioendothelioma was diagnosed. Further evaluation was performed to rule out Kasabach-Merritt syndrome.

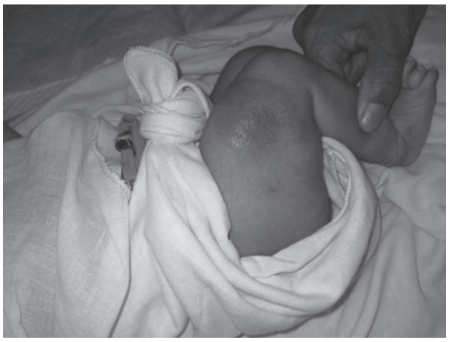

Figure 1 - Erythematous, irregular vascular lump measuring 3.8 cm x 3.6 cm observed at six hours following birth. Vitamin K injection site was seen adjacent to the vascular lump with no evidence of bleeding or involvement with the lump.

The investigations performed on day one revealed normal findings except for leukopaenia. The investigations performed on day one are presented in Table 1.

Investigations	Results	Normal range
WBC	2.6x 10^3	9,000 to 30,000/mm^3
Hb	16	12-16 g/dL
platelets	319	150 – 450 x 10^3/mm^3
PT INR APTT	15 1.3 48	14 – 17 seconds 0.9 – 1.3 28 – 48 seconds
Blood picture	No abnormalities	
Blood culture	No growth	
CRP	<2	< 5 mg/dL
USS of the lump	3.8cmX3.6cm hemangioma with two well-developed feeding arteries.	

The diagnosis was made as kaposiform hemangioendothelioma without Kasabach-Merrit syndrome based on clinical evaluation by the paediatric dermatologist together with ultrasound features of haemangioendothelioma. He was followed up to observe the clinical progression of the lesion and the possible complications. The child was followed up jointly by the paediatrician and dermatologist. Immunosuppressants were not commenced since there was no evidence of complications. The follow-up review at two weeks of age revealed gradual involution of the size of the vascular lump (Figure 2).

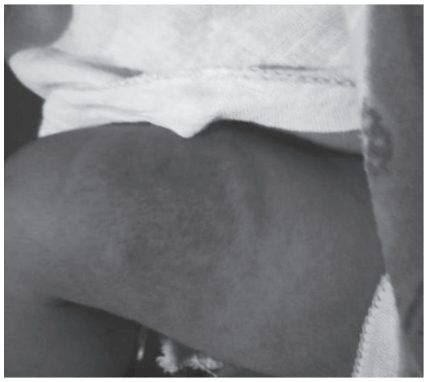

Figure 2 – Appearance of the vascular lump at two weeks of age with evidence of involution

At the eight-month follow-up, the vascular lump had completely regressed with no residual abnormalities.

Discussion

Kaposiform hemangioendothelioma is a rare locally invasive vascular tumor that can be usually seen in early childhood . Cutaneous lesions are common, infiltrating into subcutaneous tissue. Distant metastasis with visceral involvement in KHE is extremely rare and visceral lesions can be associated with Kasabach Merritt phenomenon (KMP) and lymphangiomatosis .

The aetiology of Kaposiform hemangioendothelioma is largely unknown and almost all cases appear without a specific underlying cause. Rarely, the signs and symptoms of Kaposiform hemangioendothelioma can worsen with either trauma or infections . In most children, Kaposiform hemangioendothelioma appears as a single vascular lump with an external appearance that can vary from an erythematous plaque to an indurated purple tumour. Similar to the reported child, most affected children demonstrate progressive enlargement of the vascular lump .

Complications of Kaposiform hemangioendothelioma are common necessitating the clinician to closely follow up affected children with vigilance. The reported complications include Kasabach-Merritt phenomenon , musculoskeletal disorders due to local invasion , lymphedema, and compression of vital structures .

The diagnosis of Kaposiform hemangioendothelioma is made based on clinical characteristics and ultrasound findings . Ultrasound is considered the imaging of choice14. However, the diagnosis can be delayed due to non-specific clinical findings and the lack of availability of a specific diagnostic test . Magnetic Resonance Imaging (MRI) is, however, considered the first-line assessment due to better soft tissue characterization. In this child, MRI was not performed

as ultrasound findings together with the clinical course themselves were in keeping with the diagnosis of Kaposiform hemangioendothelioma. MRI is especially recommended in the presence of unexplained thrombocytopaenia, consumption coagulopathy, and severe anaemia to rule out deep-seated Kaposiform hemangioendothelioma complicated with the Kasabach Merritt phenomenon .

Immunosuppressive therapy can be successfully used to treat Kaposiform hemangioendothelioma. Commonly used agents include prednisolone , sirolimus , and vincristine . This child was, however, managed successfully without needing specific immunosuppressive therapy as the course was uncomplicated with no evidence of Kasabach Merritt phenomenon, compressive features, or local invasion. The child was carefully followed up for eight months at which time the child was noted to have clinically improved.

Conclusion

This case report highlights the importance of considering kaposiform haemangioendothelioma as one of the differential diagnoses when a newborn presents abrupt onset rapidly enlarging vascular lump. Ruling out Kasabach-Merritt syndrome in the acute stage is crucial.

References

1. Ji Y, Chen S, Yang K, Xia C, Li L. Kaposiform hemangioendothelioma: current knowledge and future perspectives. Orphanet J Rare Dis. 2020 Feb 3;15(1):39. doi: 10.1186/s13023-020-1320-1. PMID: 32014025; PMCID: PMC6998257.

2. Adams DM, Brandão LR, Peterman CM, Gupta A, Patel M, Fishman S, et al. Vascular anomaly cases for the pediatric hematologist oncologists—An interdisciplinary review. Pediatr Blood Cancer. 2018;65(1):e26716. doi: 10.1002/pbc.26716.

3. O'Rafferty C, O'Regan GM, Irvine AD, Smith OP. Recent advances in the pathobiology and management of Kasabach-Merritt phenomenon. Brit J Haematol. 2015;171(1):38–51. doi: 10.1111/bjh.13557.

4. Mulliken JB, Anupindi S, Ezekowitz RAB, Mihm MC. Case 13-2004. New Engl J Med. 2004;350(17):1764–1775. doi: 10.1056/NEJMcpc049002.

5. Schaefer BA, Wang D, Merrow AC, Dickie BH, Adams DM. Long-term outcome for kaposiform hemangioendothelioma: a report of two cases. Pediatr Blood Cancer. 2017;64(2):284–286. doi: 10.1002/pbc.26224.

6. Drolet BA, Trenor CC, 3rd, Brandao LR, Chiu YE, Chun RH, Dasgupta R, et al. Consensus-derived practice standards plan for complicated Kaposiform hemangioendothelioma. J Pediatr. 2013;163:285–91. [PubMed] [Google Scholar]

7. Jahnel J, Lackner H, Reiterer F, Urlesberger B, Urban C. Kaposiform hemangioendothelioma with Kasabach-Merritt phenomenon: From vincristine to sirolimus. Klin Padiatr. 2012;224:395–7. [PubMed] [Google Scholar]

8. Ji Y, Chen S, Yang K, Xia C, Li L. Kaposiform hemangioendothelioma: current knowledge and future perspectives. Orphanet J Rare Dis. 2020 Feb 3;15(1):39. doi: 10.1186/s13023-020-1320-1. PMID: 32014025; PMCID: PMC6998257.

9. Chan S, Cassarino DS. Rapidly enlarging "bruise" on the Back of an infant. JAMA Dermatol. 2013;149(11):1337. doi: 10.1001/jamadermatol.2013.519.

10. Rodriguez V, Lee A, Witman PM, Anderson PA. Kasabach-Merritt Phenomenon. J Pediatr Hematol Oncol. 2009;31(7):522–526. doi: 10.1097/MPH.0b013e3181a71830.

11. Oza VS, Mamlouk MD, Hess CP, Mathes EF, Frieden IJ. Role of Sirolimus in advanced Kaposiform Hemangioendothelioma. Pediatr Dermatol. 2016;33(2):e88–e92. doi: 10.1111/pde.12787.

12. Schook CC, Mulliken JB, Fishman SJ, Alomari AI, Grant FD, Greene AK. Differential diagnosis of lower extremity enlargement in pediatric patients referred with a diagnosis of lymphedema. Plast Reconstr Surg. 2011;127(4):1571–1581. doi: 10.1097/PRS.0b013e31820a64f3.

13. Iwami D, Shimaoka S, Mochizuki I, Sakuma T. Kaposiform hemangioendothelioma of the mediastinum in a 7-month-old boy: a case report. J Pediatr Surg. 2006;41(8):1486–1488. doi: 10.1016/j.jpedsurg.2006.04.013.

14. Gong X, Ying H, Zhang Z, Wang L, Li J, Ding

A, et al. Ultrasonography and magnetic resonance imaging features of kaposiform hemangioendothelioma and tufted angioma. J Dermatol. 2019;46(10):835–842. doi: 10.1111/1346-8138.15025.

15. Ji Y, Chen S, Li L, Yang K, Xia C, Li L, et al. Kaposiform hemangioendothelioma without cutaneous involvement. J Cancer Res and Clin. 2018;144(12):2475–2484. doi: 10.1007/s00432-018-2759-5.

16. Hu P, Zhou Z. Clinical and imaging features of Kaposiform Hemangioendothelioma. Br J Radiol. 2018;91(1086):20170798. doi: 10.1259/bjr.20170798.

17. Drolet BA, Trenor CC, Brandão LR, Chiu YE, Chun RH, Dasgupta R, et al. Consensus-derived practice standards plan for complicated Kaposiform Hemangioendothelioma. J Pediatr. 2013;163(1):285–291. doi: 10.1016/j.jpeds.2013.03.080.

18. Wang C, Li Y, Xiang B, Li F, Chen S, Li L, et al. Successful Management of Pancreatic Kaposiform Hemangioendothelioma with Sirolimus. Pancreas. 2017;46(5):e39–e41. doi: 10.1097/MPA.0000000000000801.

19. Haisley-Royster C, Enjolras O, Frieden IJ, Garzon M, Lee M, Oranje A, et al. Kasabach-Merritt phenomenon: a retrospective study of treatment with vincristine. J Pediatr Hematol Oncol. 2002;24(6):459–462. doi: 10.1097/00043426-200208000-00010.

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Case Reports

Primary Mediastinal Diffuse Large B-Cell Lymphoma presenting with pyrexia of unknown origin in a 13-year-old boy

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Abstract:

Primary mediastinal large B-cell lymphoma is a rare aggressive mature B-cell lymphoma which most commonly affects adolescents and young adults. Adverse outcomes correlate with higher LDH levels (>500) and mass size over 10 cm. In this report, the authors report a teenage boy presented initially with intromittent fevers with normal haematological findings and subsequently, diagnosed to have primary mediastinal Diffuse Large B-Cell Lymphoma. This report highlights the importance of having a high index suspicion and a low threshold for further studies for haematological malignancies despite normal baseline haematological investigations so that it will prevent the diagnosis being made with delay.

Introduction

According to global data, 7% of all childhood cancers are due to Non-Hodgkin Lymphoma (NHLs)1,2. Amongst Sri Lankan children, the incidence of lymphomas and related reticuloendothelial neoplasms is 19.1 per 1000,000 male children aged 10-14 years in 20183. However, only five cases are reported in children in Sri Lanka from 0-19 years with a diagnosis of Diffuse Large B-Cell Lymphoma (DLBCL)1. Childhood DLBCLs responds well to muti-drug chemotherapy and Event Free Survival (EFS) is around 90%. Here we present a 13-year-old boy from Western province of Sri Lanka who presented with moderate episodic fever for 4 months in association with loss of appetite and weight and subsequently, was diagnosed to have DBLCL.

Case report

The index child is a 13-year-old boy who was admitted with a history of episodic fever for 4 months. Fever was documented at 100 – 101 F and associated with chills but not rigors. Fever was intermittent, spanning throughout the day. Child was well despite fever. Apart from mild headache, there was no photophobia, phono phobia, nausea or vomiting. No behavioral changes were noted. There were no respiratory, urinary or gastro-intestinal symptoms to account for fever. Fever episodes were 4 to 5 days in duration and settled spontaneously without any treatment in most occasions. The child also reported having loss of appetite and has lost 5 kg of weight over last 4 months. There was no significant travel history, tick bites or exposure to tuberculosis. Past medical and drug history were insignificant. There was no family history of malignancies.

Clinical examination was insignificant with no lymphadenopathy, pallor, icterus or hepatosplenomegaly. His Full Blood Count (FBC) was also normal and no abnormal cells were present in Blood Picture. Moderately elevated ESR (76 mm 1st hour) and high CRP (56 mg/dL) were present. His chest roentgenogram revealed a mediastinal mass which was radiologically confirmed as a mediastinal lymphoma. Contrast enhanced CT chest and abdomen revealed multiple enlarged mediastinal lymph nodes involving 2, 3, and 6 mediastinal lymph node groups. Left supraclavicular lymph node was also enlarged and prominent mesenteric lymph nodes were visualised in CECT abdomen. The appearances are suggestive of lymphoma. Histological diagnosis of DLBCL was made subsequently by ultrasound guided left supraclavicular lymph node biopsy.

His investigations results were as follow (Figure 1,2 and 3).

Figure 1 – The laboratory investigation results

Investigations		
Total WBC	**5.91**	(4.5-10.5)
Neutrophils	**3.787**	(1.8-7.3)
Lymphocytes	**1.68**	(0.9-4.7)
Eosinophils	**0.05**	(0.45-0.84)
Hemoglobin	**10.4**	(13.0 – 16.5)
PCV	**32.4**	(39 – 47)
MCV	**80.5**	(76 – 96)
MCH	**25.8**	(27 – 32)
Platelets	**503**	(150 – 450)
CRP	**91**	(0 – 5)
ESR	**58**	(<10)
Sodium	**144**	(137 – 145)
Potassium	**5.0**	(3.5 – 5.1)
AST	**22**	(0-35)
ALT	**29**	(0-40)
Creatinine	**42**	(70-115)
Urea	**15**	(8-50)
LDH	**248**	(210 - 240)
Blood picture	No abnormal cells.	

Figure 2 - Posteroanterior view of chest roentgenogram showing a prominent mediastinal mass towards the left hemithorax s compressing the lower trachea and the left main bronchus.

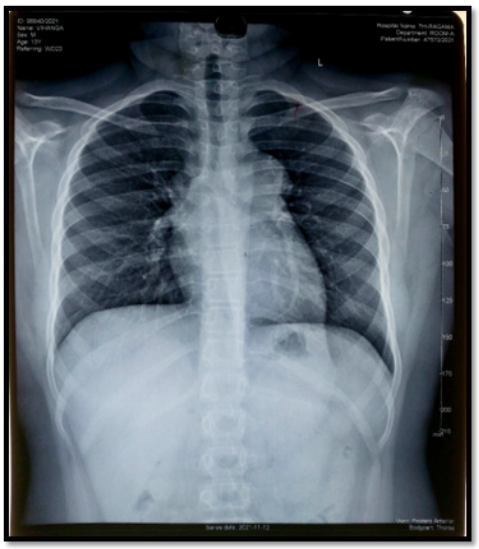
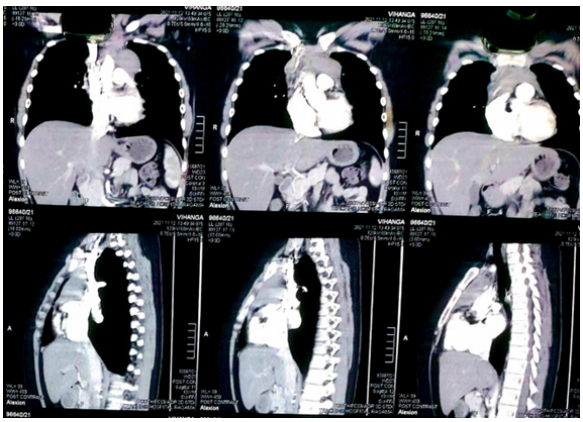


Figure 3 - CECT Chest and Abdomen showing mediastinal lymph node mass involving 2, 3, and 6 mediastinal lymph node groups. Left supraclavicular lymph node was enlarged and prominent mesenteric lymph nodes were visualised.



Discussion

Lymphomas are a malignancy of the lymphatic system. There two main types lymphoma are Hodgkin and Non-Hodgkin Lymphomas. According to global data, 7% of all childhood cancers are due to Non-Hodgkin Lymphoma (NHLs)1,2. Around 800 new children are diagnosed with NHLs every year in United States4,5. It was 2-3 times commoner in boys. These statistics are lower in Sri Lanka and male predominance is statistically lesser compared to western countries3.

A Sri Lankan study assessed the pattern of World Health Organization (WHO) sub types of lymphoma in a sample of adult patients6. In adults, the overall outcome of DLBCLs is poorer and very heterogeneous. However, the childhood DLBCLs respond well to muti-drug chemotherapy and Event Free Survival is around 90%. Earlier the diagnosis is made, better the prognosis.

The reported child had symptoms for over 4-5 months. However, the symptoms were non-specific and absence of any haematological abnormalities lead to a delayed diagnosis. Physical examination did not provide any clue about the diagnosis as lymphadenopathy and hepatosplenomegaly were absent. Similarly, his haematological parameters and blood picture were normal. Despite lack of findings in history, physical examination and haematological assessment, a high degree of clinical suspicion prompted the authors perform further studies that established the final diagnosis of DLBCL.

Conclusion

This case report highlights the fact that a serious illness like NHLs can present with innocent symptoms and clinical examination can be completely normal. Hematological indices also can be normal. It is therefore very important to have a high index suspicion and a low threshold for further studies that will prevent the diagnosis being made with delay.

List of abbreviations

CECT – Contrast Enhanced Computed Tomography
NHLs – Non Hodgkin Lymphomas
DLBCL – Diffuse Large B cell Lymphoma
CBC – Complete Blood Count

References

1. Swerdlow SH, Campo E, Harris NL, Jaffe ES, Pilleri SA, Stein H, Thiele J, Vardiman JW (eds). World Health Organization classification of tumours of haematopoietic and lymphoid tissue. Lyon: IARC Press, 2008

2. Morton LM. Dissecting lymphoma incidence to inform epidemiologic and clinical research. Leukemia & Lymphoma 2013; 54: 1575-76.

3. Childhood Cancer Registry in Sri Lanka. Available from: https://www.nccp.health.gov.lk/storage/post/pdfs/Childhood%20Cancer%20Registry%20Sri%20Lanka.pdf

4. Clarke CA, Glaser SL. Changing incidence of non Hodgkin lymphomas in the United States. Cancer 2002; 94: 2015-23

5. Miles RR, Raphael M, McCarthy K, et al. Pediatric diffuse large B-cell lymphoma demonstrates a high proliferation index, frequent c-Myc protein expression, and a high incidence of germinal center subtype: Report of the French-American-British (FAB) international study group. Pediatr Blood Cancer. 2008

6. 6. Waravita TS, Wijetunge TS, Ratnatunga NV. Pattern of lymphoma subtypes in a cohort of Sri Lankan patients. Ceylon Med J. 2015 Mar;60(1):13-7. doi: 10.4038/cmj.v60i1.7143. PMID: 25804912.

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Case Reports

Two cases of "Rusty pipe syndrome"; a potential barrier for establishment of breastfeeding.

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Abstract:

Rusty Pipe syndrome is a rare, benign, self-limiting condition which presents with blood-stained nipple discharge during the period of establishment of breastfeeding. Since the presentation can cause a significant anxiety in the mother, establishment of breastfeeding can be adversely affected. We report mothers of two newborn babies who had brownish breast milk in the absence of a known predisposing factor such as cracked nipples, mastitis or breast infection. Both mothers were managed with lactation support and breastfeeding counselling and were able the establish breastfeeding successfully. This report highlights the importance of accurate recognition of this benign condition and assisting mothers to overcome anxieties and establish breastfeeding successfully.

Introduction

Rusty pipe syndrome is a rare phenomenon which is characterised by painless, blood-stained nipple discharge during the first few days following delivery. The appearance of breast milk in the presence of this condition can be pink, orange, brown or rusty coloured. The blood-stained discharge can be caused by a number of factors which include cracked nipples, trauma, mastitis, and intraductal papilloma . The term "rusty pipe syndrome" is coined as the breast milk looks similar to dirty water coming out of an old rusty pipe. Rusty pipe syndrome can potentially cause a significant anxiety in the mother and may adversely affect establishment of breastfeeding. The authors report two cases of "Rusty pipe syndrome" and it is highly important to awareness of this rare and benign condition in order to help mothers to establish breastfeeding successfully.

Case 1

A 32-year-old primigravida delivered a baby girl with a birth weight of 2.9 kg following an uncomplicated antenatal period. The delivery was made by elective lower-segment caesarean section as per mother's request. Her breast milk was noted to be brownish in colour on day 1 and she was anxious about the appearance of the breast milk (Figure 1). The brownish discolouration of breast milk was evident in milk expressed by both breasts. She was assisted by the lactation management team to establish breastfeeding. There was no history feeding difficulties at birth including cracked nipples, inverted nipples or evidence of breast infection. The mother also denied any history of trauma to breasts. The breast examination did not reveal any evidence of ulcers, cracks, or fissures. Newborn examination did not reveal any natal teeth. Ultrasound breast was normal.

Over the next 48 hours, the colour of breast milk gradually became light brownish and by day four, the colour of breast milk was normalised with no active intervention. Currently, the baby is two weeks old and breast feeding is well established. The neonate showed a normal pattern of weight gain.

Figure 1 – The appearance of breast milk on day 1

Case 2

A 26-year-old primiparous mother at 29 5/7weeks of period of gestation was transferred from a base hospital with labour pains. Twin babies were delivered vaginally following admission. Both babies were taken to Neonatal Intensive Care Unit due to very prematurity and low birth weight. In the antenatal period, mother was followed by a maternity clinic regularly and no health problems were identified. For initiation of breastfeeding, the mother was visited by neonatal nurse in the first hour and showed her hand expressing of the milk. Brownish milk from both breasts were noted (Figure 2).

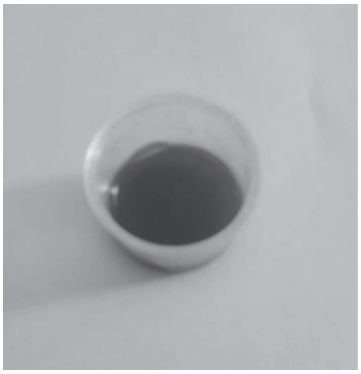

Figure 2 – The appearance of breast milk while being expressed

Figure 3 – The appearance of breast milk as compared to normal breast milk and 0.9% saline

The mother denied any history of pain, trauma, or recent infection. Examination of the breasts revealed no engorgement, tenderness, and erythema or mass lesion. The nipples and areola had no erosions, ulcers, or cracks. A breast ultrasound was performed and the findings were normal. No solid nodular images were found. Cytological analysis of milk was arranged. No abnormal or malignant cells were noted. The mother was reassured, so that breastfeeding could be continued. The breast milk improved to normal colour on Day 5.

Discussion

Rusty pipe syndrome is a painless self-limiting condition . It is noticed as discoloured breast milk in pregnant women in the third trimester or within the first days of breastfeeding . The condition is neglected unless the mother is expressing the milk. It is commonly bilateral. It is thought to be due to the delicate network of capillaries, which get traumatized easily and result in blood leaking into breast secretions . Diagnosis of rusty pipe syndrome is made by normal clinical examination of the breast and excluding the sinister causes like ductal papilloma by imaging and cytology analysis1. Continuation of breastfeeding is advocated as the condition is self-limited, and most decoloured breast milk turns to normal colour within the first week. Blood contents in breast milk may result in vomiting or regurgitation in the newborn in some cases5. Understanding about rusty pipe syndrome among health care professionals would be very helpful to relieve anxiety in nursing mothers. Avoidance of unneeded interventions, and maintaining exclusive breastfeeding must be the goal of lactation support and breastfeeding counselling. Mothers should also be informed that it may recur in their future pregnancies as well.

If the blood-stained nipple discharge persists, this should be further investigated with ultrasonography and by detailed clinical evaluation . If the ultrasonography reveals an abnormal lesion, further investigation is warranted with mammography. However, most mothers with “Rusty pipe syndrome” will start having normal colour breast milk within the first few days and investigations are unnecessary.

Conclusion

Rusty pipe syndrome is a rare, harmless condition in which the breast milk appears reddish or brownish in colour during the first few days of the life of the newborn. It is important that practitioners caring for lactating mothers are well-aware of this rare condition as discoloured breast milk can cause significant anxiety in the mother. Lactation support and breastfeeding counselling help mothers to establish breastfeeding successfully.

References

1. Deboni FA, Moldenhauer M, do-Nascimento MB. “Rusty pipe” syndrome: benign and rare cause of bloody nipple discharge during breastfeeding: case report. Residencia Pediatrica. 2018;8:151–

2. Gueye M, Kane-Gueye SM, Mbaye M, Ndiaye-Gueye MD, Faye-Dieme ME, Diouf AA, et al. Rusty pipe syndrome in a 22-year-old primigravida at 26 weeks' gestation. S Afr J Obstet Gynaecol. 2013;19:17–8.

3. Bahar Kural and Serap Sapmaz Breastfeeding medicine Volume 15, Number 8, 2020 Mary Ann Liebert, Inc. DOI: 10.1089/bfm.2020.0055

4. Faridi MM, Dewan P, Batra P. Rusty pipe syndrome: Counselling a key intervention. Breastfeed Rev 2013;21:27–30.

5. Mehmet Nevzat Cizmeci, Mehmet Kenan Kanburoglu Rusty-Pipe Syndrome: A Rare Cause of Change in the Color of Breastmilk Breastfeeding medicine Volume 8, Number 3, 2013 ª Mary Ann Liebert, Inc. DOI: 10.1089/bfm.2012.0143

6. Mitchell KB, Johnson HM, Eglash A, Academy of Breastfeeding Medicine ABM clinical protocol #30: breast masses, breast complaints, and diagnostic breast imaging in the lactating woman. Breastfeed Med. 2019;14:208–14.

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Case Reports

Intestinal obstruction caused by endometriotic deposits: A case report

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Abstract:

Introduction: Endometriosis is a benign gynecological condition affecting women of reproductive age. The condition principally presents with symptoms like dysmenorrhea, pelvic pain and subfertility. Although endometriosis in the gastrointestinal tract is not an uncommon occurrence, it causing acute intestinal obstruction has been seldom reported in literature.
Case report: We present a case history of a 40 year old woman, who had presented with acute intestinal obstruction. A sigmoid colectomy was done for suspected malignant obstruction of rectosigmoid junction. Histology revealed endometriotic tissue deposits.
Discussion: Incidence of endometrial tissue in the gastrointestinal tract greatly varies according to the site. Greatest incidence is reported to be in the sigmoid colon and rectum. Acute intestinal obstruction can infrequently occur due to endometriosis. Clinical diagnosis in such a case can be challenging, so is radiological confirmation. A combination of medical and surgical therapies can be attempted for endometriosis. In emergency presentation surgery plays a pivotal role in relieving the obstruction be it a Hartmann's procedure or placement of a stent.
Conclusion: Intestinal endometriosis should be considered a differential diagnosis in women of reproductive age, presenting with intestinal obstruction, which requires a high index of suspicion where relevant, given the history.

Introduction

Endometriosis is a benign gynaecological condition affecting around 10% of women of reproductive age across the world(1). The cumulative prevalence of endometriosis is around 6% according to I.J.Rowlands, in Australia(2). A similar figure of 6.8% has been found as the prevalence rate of endometriosis in East Asia in literature(3). Women usually complain of dysmenorrhea, pelvic pain or subfertility when the endometriosis involves the pelvic peritoneum, which is the case in majority of this condition. But the symptoms can vary depending on the site of deposition of the tissue. Gastrointestinal tract is the most common extra pelvic site of endometriosis affecting around 5%-12% of those with endometriosis(4). Though an acute presentation is uncommon with gastrointestinal tract involvement, rectosigmoidal endometriosis can present as an acute abdomen due to its narrowest intraluminal diameter in the alimentary tract. We present such a case of a woman who has presented with bowel obstruction due to an endometriotic lesion in the rectosigmoid junction.

Case report

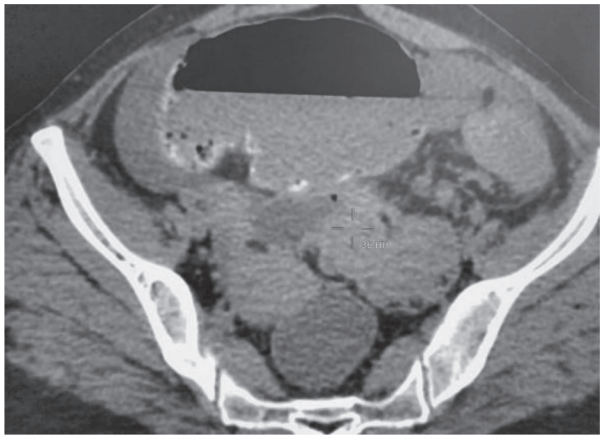
A 40 year old woman presented to the surgical casualty with diffuse abdominal pain associated with absolute constipation and distension of abdomen for two days.

The patient began experiencing dysmenorrhea 3 years ago. The pain started on the first day of menstruation and has not outlasted periods. She also complained of nausea and diarrhea during this time. Denied heavy menstrual bleeding or irregular bleeding. She's nulliparous and has had regular menses since menarche. Evaluation by a Gynaecologist has revealed fibroids 6 months earlier. Her past medical and surgical history were unremarkable.

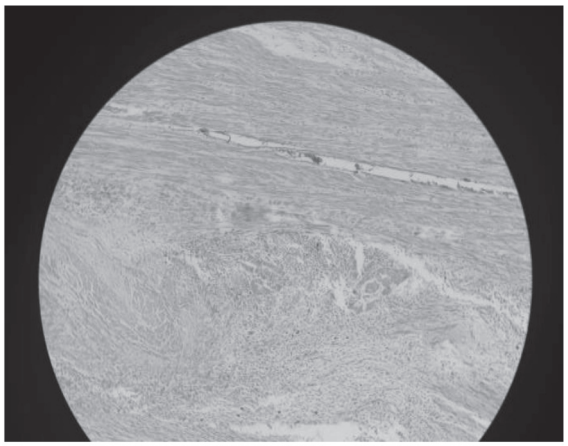
Two months ago she started experiencing episodes of colicky type of pain in the left iliac fossa. She had multiple episodes throughout the day each lasting for 15-20 minutes. She complained of multiple diarrheal episodes during the same time period. Two days prior to admission her pain worsened progressively and persisted throughout the day. Had

vomited in large amounts but denied passage of stools or flatus. She has noted progressive abdominal distension during the same period of time. Had one episode of bleeding per rectum following admission. Physical examination was significant for generalized abdominal tenderness and distension. No abdominal masses or free fluid noted. Digital rectal examination revealed bleeding per rectum. A blood pressure of 110/70 mmHg and pulse rate of 78 bpm were recorded on admission.

All the blood investigations turned out to be normal. Abdominal X-ray showed dilated bowel loops. Erect chest X-ray had no gas under diaphragm. Ultrasound scan of abdomen revealed moderate amount of fluid in pelvic cavity and hepatorenal pouch. NCCT abdomen and pelvis evidenced a hyperdense soft tissue lesion measuring 3.9x4.5cm in size in sigmoid colon causing significant luminal narrowing leading to intestinal obstruction.



Suspected lesion at rectosigmoid junction mimicking a malignancy.



Bland endometrial gland with stroma within muscularis propria

An emergency explorative laparotomy was carried out leading to a finding of grossly dilated caecum with mucosal tear and impending rupture. A near total obstructing mass lesion was found at distal sigmoid colon just proximal to rectosigmoid junction. Few enlarged lymph nodes were noted along sigmoid vessels. Hartmann's procedure performed removing the proximal rectum and sigmoid colon. Specimen was sent for histological analysis. The post-operative course was uneventful.

Histology of the specimen showed endometriosis in sigmoid colon. On sectioning through the bowel no tumor was visible. But multiple areas of ulceration and small polypoidal lesions were noted. Microscopy showed endometrial glands embedded in stroma in muscularis propria and serosa of bowel. Areas of hemorrhage and fibrosis were noted.

Discussion

Endometriosis in gastrointestinal tract has been reported in quite a significant number of cases. The reported incidence of the involvement of different intestinal sites varies greatly in the literature, with the rectosigmoid colon, small bowel, appendix and caecum affected in 50-90%, 2-16%, 3-18% and 2-5% of cases respectively(5). Endometriotic tissue in large bowel present with bleeding per rectum, dyschezia and intestinal obstruction. Small bowel endometriosis may manifest with nonspecific symptoms such as abdominal pain and bloating. Sigmoid colon and rectum are the most commonly involved sites in a woman with endometriosis(6). The etiology of endometriosis remains unknown and controversial. There are many theories but currently the most widely accepted theory is that of 'retrograde menstruation' causing the implantation and growth of endometriosis on the serosal surface of extra uterine organs or occurring secondary to metaplasia in the pelvic peritoneum(7–11).

In our case, though the patient had a 3 year history of dysmenorrhea, she has not complained of suggestible symptoms of intestinal endometriosis. Her presentation with acute abdomen, and an obstructive lesion seen at surgery were attributed to a neoplastic growth at the outset. The histology specimen was reported to be a sigmoid colon endometriosis and it excluded a malignancy.

Although the most common place of endometriosis in the gastrointestinal tract is rectosigmoid junction, acute abdomen has been rarely reported(5). Around 1% of these cases have been complicated with intestinal obstruction requiring surgical

intervention(12). Clinical features and transvaginal ultrasound scan suggest an endometriosis but the definitive diagnosis is made on histological report. MRI has been proven to be sensitive for diagnosis of intestinal endometriosis. Management depends on individual cases and preferences. Medical treatment with hormonal therapy such as OCP, Danazol or Gonatrophin antagonists can be attempted as first line therapy for intestinal disease when there is no obstruction. This remains controversial as there are only few reported cases of medical therapy being successful. But hormonal therapy was equally efficient in improving pain compared to surgery and was associated with lower complication rates in women with rectosigmoid endometriosis(13). It is argued by some that the rare but potential risk of malignant transformation makes surgical resection mandatory(14). When the surgery is elective then a laparoscopic approach should be favored although it is important to explain the potential complications such as rectovaginal fistulae(15,16). Surgery is absolutely indicated in acute or sub-acute bowel obstruction that fails to resolve as well as in endometriotic tumors or when it is impossible to exclude a malignancy(17,18).In an emergency setting, the main aim of surgery should be to relieve the obstruction. The treatment of choice is usually an emergency procedure (either Hartmann procedure or resection and anastomosis with stoma placement). This approach entails all the risks related to emergency procedures and can have important psychological and biological drawbacks. Endoscopic prosthesis placement as bridge to surgery is a feasible therapeutic strategy in colonic obstruction due to endometriosis. It brings about all the advantages of an expedited one step laparoscopic surgical procedure(12). If endometriosis is suspected intra-operatively, then as many ectopic deposits as possible should be excised. Endometriosis has a recurrence rate of up to 89% after surgery(19). The rate increases with incomplete resection of tissues.

Conclusion

Although endometriosis is rare to cause bowel obstruction, surgeons should consider it as a differential diagnosis in women of reproductive age. A large suspicious mass at surgery should be dealt as one of malignant growth.

Reference

1. Blamble T, Dickerson L. Recognizing and treating endometriosis. J Am Acad Physician Assist. 2021;34(6).
2. Rowlands IJ, Abbott JA, Montgomery GW, Hockey R, Rogers P, Mishra GD. Prevalence and incidence of endometriosis in Australian women: a data linkage cohort study. BJOG An Int J Obstet Gynaecol. 2021;128(4).
3. Yen CF, Kim MR, Lee CL. Epidemiologic factors associated with endometriosis in East Asia. Vol. 8, Gynecology and Minimally Invasive Therapy. 2019.
4. Mota LM, Hirsch BS, Seligman R. Large Bowel Endometriosis Mimicking Colorectal Cancer. Case Rep Med. 2020;2020.
5. Teke Z, Aytekin FO, Atalay AO, Demirkan NC. Crohn's disease complicated by multiple stenoses and internal fistulas clinically mimicking small bowel endometriosis. World J Gastroenterol. 2008;14(1).
6. Miller LS, Barbarevech C, Friedman LS. Less frequent causes of lower gastrointestinal bleeding. Vol. 23, Gastroenterology Clinics of North America. 1994.
7. Scarmato VJ, Levine MS, Herlinger H, Wickstrom M, Furth EE, Tureck RW. Ileal endometriosis: Radiographic findings in five cases. Radiology. 2000;214(2).
8. Siristatidis C. What have the 'omics done for endometriosis? Vol. 15, Medical Science Monitor. 2009.
9. Popoutchi P, dos Reis Lemos CR, Silva JCR, Nogueira AA, Feres O, da Rocha JJR. Postmenopausal intestinal obstructive endometriosis: Case report and review of the literature. Sao Paulo Med J. 2008;126(3).
10. Szucs RA, Turner MA. Gastrointestinal Tract Involvement by Gynecologic Diseases. Radiographics. 1996;16(6).
11. De Ceglie A, Bilardi C, Blanchi S, Picasso M, Di Muzio M, Trimarchi A, et al. Acute small bowel obstruction caused by endometriosis: A case report and review of the literature. World J Gastroenterol. 2008;14(21).

12. Calcagno P, Viti M, Cornelli A, Galli D, D'Urbano C. Intestinal obstruction caused by endometriosis: Endoscopic stenting and expedited laparoscopic resection avoiding stoma. A case report and review of the literature. Int J Surg Case Rep [Internet]. 2018;44:75–7. Available from: https://doi.org/10.1016/j.ijscr.2018.02.012

13. Andres MP, Mendes RFP, Hernandes C, Araújo SEA, Podgaec S. Hormone treatment as first line therapy is safe and relieves pelvic pain in women with bowel endometriosis. Einstein (Sao Paulo). 2019;17(2).

14. Bianchi A, Pulido L, Espín F, Hidalgo LA, Heredia A, Fantova MJ, et al. Endometriosis intestinal. Estado actual. Vol. 81, Cirugia Espanola. 2007.

15. Dubernard G, Piketty M, Rouzier R, Houry S, Bazot M, Darai E. Quality of life after laparoscopic colorectal resection for endometriosis. Hum Reprod. 2006;21(5).

16. Duepree HJ, Senagore AJ, Delaney CP, Marcello PW, Brady KM, Falcone T. Laparoscopic resection of deep pelvic endometriosis with rectosigmoid involvement. J Am Coll Surg. 2002;195(6).

17. Denève E, Maillet O, Blanc P, Fabre J-M, Nocca D. [Ileocecal intussusception secondary to a cecal endometriosis]. J Gynecol Obs Biol Reprod. 2008;37(8).

18. Garg NK, Bagul NB, Doughan S, Rowe PH. Intestinal endometriosis-A rare cause of colonic perforation. World J Gastroenterol. 2009;15(5).

19. Ceccaroni M, Bounous VE, Clarizia R, Mautone D, Mabrouk M. Recurrent endometriosis: a battle against an unknown enemy. Vol. 24, European Journal of Contraception and Reproductive Health Care. 2019.